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Original Communications

THE EFFECT OF PREOPERATIVE STREPTOMYCIN ON THE INCIDENCE OF BRONCHOPLEURAL FISTULA AFTER PULMONARY EXCISION FOR TUBERCULOSIS

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THE aim of this communication is to show that after pulmonary excision for tuberculosis, tuberculous lesions remaining in the bronchial stump contribute to the development of bronchopleural fistula and that streptomycin, by controlling this infection *before* operation, reduces the incidence of this formidable postoperative complication.

In order to assess the influence of bronchial disease on the complications following pulmonary excision, it is necessary to eliminate other factors which may affect the incidence of these complications. Because thoracoplasty, phrenic interruption, and pneumoperitoneum may help to prevent bronchopleural fistula, empyema, and reactivation of residual tuberculosis, no patient was included in this study who had any of these operations performed before, at the time of, or after pulmonary excision. In all cases an essentially similar operative technique was used. Some patients were not given streptomycin before operation but all patients received streptomycin for at least four weeks after the excision. One gram of streptomycin was given daily. All patients were observed for at least three months after operation; the average period of observation was nine months.

The relationship between postoperative complications and the site of bronchial division was studied in 140 patients. It was found that there was no significant difference in the two sides but the incidence of complications did vary with the part of the lung removed. Table I shows that after excision of the lung, or all or part of the upper lobe, fistula, empyema, and spread of disease occurred; but after removal of all or part of the middle or lower lobe neither fistula nor empyema developed although some spreads occurred. However, within the group developing fistula, its incidence did not vary significantly with the part excised.

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TABLE I. COMPLICATIONS AFTER PULMONARY EXCISION FOR TUBERCULOSIS

PART REMOVED	TOTAL NO. OF PATIENTS	WITH COMPLICATIONS		
		FISTULA	EMPYEMA	SPREAD
Lung, or all or part of upper lobe	107	27	2	4
All or part of middle or lower lobe	33	0	0	4

Since fistula did not develop unless all or part of the upper lobe was removed, the effect of preoperative streptomycin on the incidence of fistula was observed after excision of lungs and all or part of the upper lobes only. Thirty-three pneumonectomies, forty-six upper lobectomies, and twenty-eight upper segmental excisions were performed on 107 patients. Of these patients, thirty-five had para-aminosalicylic acid with the streptomycin and seventeen had received streptomycin previously but more than six months before operation. In Table II the patients are grouped according to whether they received preoperative streptomycin or not. In the group which did not receive streptomycin before operation 50 per cent developed fistulas. Whereas in the group which received streptomycin in the immediate preoperative period 16 per cent developed fistulas.

TABLE II. EFFECT OF PREOPERATIVE STREPTOMYCIN TREATMENT ON INCIDENCE OF FISTULA

	TOTAL	WITH FISTULAS
Patients not receiving streptomycin before operation	28	14 (50%)
Patients receiving some streptomycin before operation	79	13 (16%)

The central end of the divided bronchus was examined for evidence of tuberculosis in thirty-one operation specimens (seventeen lungs, ten lobes, and four segments excised for this disease). Bronchi considered to be tuberculous showed typical follicles of lymphocytes, epithelioid cells, and multinucleated giant cells; in some follicles there was caseation. Although they occasionally extended more deeply, these lesions were found always in the submucosa. In some cases the overlying epithelium was intact, in others it was ulcerated by the subepithelial infection. These tuberculous lesions were found in twenty-seven of the thirty-one bronchi sectioned. The British Tuberculosis Association[1] reported that of ninety-four operation specimens, histologic evidence of tuberculosis was found at the point of bronchial transection in 95 per cent. The 87 per cent of the present smaller series agrees closely with this incidence (Table III).

TABLE III. INCIDENCE OF BRONCHIAL TUBERCULOSIS

AUTHOR	NO. OF OPERATION SPECIMENS	TUBERCLULOSIS IN TRANSECTED BRONCHUS
British Tuberculosis Association	94	95%
Present series	31	87%

Since tuberculosis is very common in the central end of the bronchi of excised specimens, it is highly probable that tuberculous lesions are present in

the bronchial stumps left in the patients. In two patients who had upper lobectomies, no evidence of tuberculosis was found in the divided bronchi of the operation specimens. Both patients developed bronchopleural fistulas after operation. After completion pleuropneumonectomy in one and at necropsy in the other, tuberculous lesions were found in the bronchi at the site of the fistulas.

Tuberculous infection of the bronchial stump might cause the disease to spread to residual lung tissue or the pleural cavity. It is likely also to interfere with healing of the divided bronchus and lead to the development of a bronchopleural fistula. Thus it seems reasonable to suppose that the reduced incidence of fistula shown in Table II was due to suppression of bronchial infection by the preoperative streptomycin. Furthermore, Table IV shows that the incidence of fistula is influenced by the duration of streptomycin treatment before operation. In the group of twenty-eight patients who did not receive the benefit of streptomycin given in the immediate preoperative period one-half developed fistulas. When streptomycin was given to forty-five patients for three weeks or less before operation the incidence of fistula was reduced to one-fourth; of thirty-four patients in whom streptomycin treatment was maintained for more than three weeks only two developed fistulas.

TABLE IV. INFLUENCE OF DURATION OF PREOPERATIVE STREPTOMYCIN TREATMENT ON INCIDENCE OF FISTULA

PREOPERATIVE STREPTOMYCIN	TOTAL NO. OF PATIENTS	WITH FISTULAS
None	28	14 (50%)
3 weeks or less	45	11 (24%)
More than 3 weeks	34	2 (6%)

SUMMARY

Tuberculous lesions are very common in bronchial stumps and probably are an important factor in the development of bronchopleural fistula. This view is supported by the finding that preoperative streptomycin treatment reduced the incidence of fistula after excision of the lung or all or part of the upper lobe. It is evident also that the patient may not be protected adequately by streptomycin given for a short period before operation and it is advocated that pulmonary excision for tuberculosis be preceded by streptomycin treatment for at least three weeks.

Patients were included in this study with the kind permission of several physicians and surgeons at St. George's Hospital, London, Poole Hospital, Middlesbrough, Milford Chest Hospital, Milford, Grove Park Hospital, London, and King George V Hospital for Diseases of the Chest, Godalming.

REFERENCE

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SIMULTANEOUS OCCURRENCE OF SIMPLE AND MALIGNANT TUMORS IN THE ESOPHAGUS

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CARCINOMA of the esophagus is a common cause of dysphagia in older people. Simple tumors of the esophagus are less often responsible, and of these leiomyoma is probably the most frequent type. There has been a renewed interest in nonmalignant conditions causing obstruction in the last few years (Moersch and Harrington,[4] Calmenson and Clagett,[2] Myers and Bradshaw[5]), but a search of the literature does not reveal any case where the two conditions occur simultaneously in the same patient. The case report presented appears to be unique in this respect.

CASE REPORT

A married male patient (a builder), aged 54 years, was admitted to the hospital on Oct. 1, 1952, with the following history:

History.—Various aches and pains had been felt across the chest for twelve months and were put down to "indigestion," although they occurred at any time of the day. Three months previously the patient had noticed that cold water regurgitated if he drank too fast, and over the next few days he felt food sticking as he swallowed. This was accompanied by pains in the epigastrium. The amount of vomiting varied from day to day. He thought he was losing weight. This patient ordinarily had a good appetite, but had been living on fluids for the past six weeks.

He had had measles and chicken pox as a child. He had received superficial wounds in World War I. In 1934 he had undergone appendectomy. One brother had died of cancer of the neck(?).

Personal Habits.—He smokes one-half ounce of tobacco a day and drinks two pints of beer on week ends.

Examination.—The patient was a thin, edentulous subject. He was not anemic. There was no clubbing. A few small lymph nodes were palpable in left axilla; otherwise there were no abnormal physical findings.

Investigations.—His normal weight was 152 pounds (in clothing). On admission he weighed 126 pounds (without clothing). He was apyrexial. Vital capacity was 3,850 ml. Hemoglobin was 113 per cent 100 = 14.8 Gm. White blood cells were 11,300 per cubic millimeter; polymorphs, 64 per cent; lymphocytes, 28 per cent; eosinophils, 3 per cent; and monocytes, 5 per cent. The patient's blood group was A, Rh positive. There was normal flora in the sputum. Blood urea was 36 mg. per 100 ml.; total serum protein, 6 Gm. per 100 ml.; albumin, 3.5 Gm. per 100 ml.; and globulin, 2.5 Gm. per 100 ml. Electrocardiogram showed right ventricular preponderance only in Leads I, II, and III. Chest x-ray showed no abnormality apart from emphysema; the heart was small; barium swallow showed an esophageal stricture at the level of the bifurcation of the trachea.

Esophagoscopy (Oct. 6, 1952).—Neoplastic stricture present at 30 cm. from the teeth was reported to be histologically a squamous cell carcinoma.

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From the Institute of the Diseases of the Chest, London, England.

Operation.—Esophagectomy (Mr. D. Barlow; Dr. Leighton), a two-stage operation, was performed at one session.

1. *Abdominal part:* A left upper paramedian incision was made. Findings: No obvious malignant spread, although the glands in the lesser omentum toward the cardia were freely palpable and firmer than normal. Specimen was sent for section (the report of which showed no involvement by growth). The stomach was mobilized from the duodenum to the lower end of the esophagus. The abdomen was closed and the patient was turned onto the right side.

Fig. 1.—Barium swallow. There is a short stenosis, with dilatation visible above it. On the right (left of x-ray) a smooth, rounded projection 1.25 cm. in length is present below the stenosis.

2. *Right thoracotomy:* This was done through the bed of the seventh rib, which was resected. Findings: A malignant stricture of the esophagus with secondary malignant nodules palpable in the region of the thoracic inlet adherent to both trachea and esophagus. Procedure: The segment containing the stricture was removed after mobilization of the esophagus from below. The cardiac orifice of the stomach was closed and invaginated. End-to-side anastomosis was performed. The level of the transection of the esophagus was 1½ inches (3.75 cm.) beneath the first rib. The chest was closed, with drainage over penicillin tablets.

The patient soon began swallowing soft foods without difficulty or discomfort, until Oct. 23, 1952, when he complained of dyspnea. On examination, numerous coarse moist sounds were present in all areas of the chest. X-ray showed air and fluid in the pleural

cavity. No lung was visible. Aspiration yielded 7 ounces of honey-colored, odorless fluid, culture of which yielded a growth of *Staphylococcus aureus* and gram-negative bacilli of coliform type, both sensitive to Chloromycetin and Aureomycin. An intercostal drain was inserted to an underwater seal. By Oct. 31, 1952, expansion of underlying lung was almost complete, and the tube was removed. In the next four weeks, the patient had intermittent trouble with swallowing, with occasional vomiting. The chest infection gradually resolved. He was discharged from the hospital on Nov. 22, 1952. On Jan. 20, 1953, he died. No post-mortem examination was made.

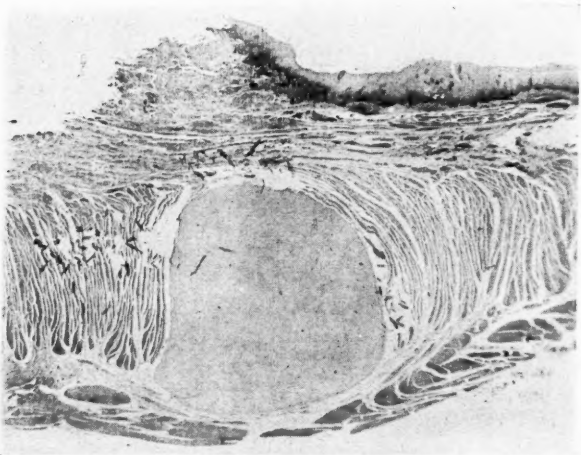

Fig. 2.—Longitudinal section of the esophagus showing intramuscular leiomyoma, with overlying carcinoma.

Pathologic Report of Specimen Removed (Dr. K. W. Hinson).—"A portion of the esophagus with a carcinomatous ulcer in the centre. Immediately distal to this ulcer there is a small roughly spherical tumour, bulging externally. The carcinoma is of the squamous celled type and has metastasized to the attached lymph node. The staining reactions of the second tumour are indefinite. It consists of interlacing whorls of elongated cells, and is probably a benign neurilemmoma."

Examination of further sections showed the carcinoma to be a typical squamous example. It had metastasized through the muscular coat, and several lymphatic vessels were permeated by the growth. The second tumor lying in the circular muscle coat opposite the carcinoma, is discrete, but not truly encapsulated. Its edges fade into the adjacent muscle tissue.

Low-power view shows that it consists of many interlacing fibrils, with scattered nuclei among them. The fibrils take up hematoxylin and eosin stain less readily than do the myofibrils. No nerve fibers are visible within the tumor, and Masson's trichrome stain is negative for nerve axons. Only a few blood vessels are present.

High-power view (×360) shows the tumor to consist of a mixture of smooth muscle and collagenous fibrils. In places, a foamy and myxomatous appearance is present. A few plasma cells are seen. The nuclei are spindle-shaped and their staining reaction fairly homogeneous throughout the section. Very few mitotic figures are visible. Palisading of nuclei is noticeably absent.

Conclusion.—The tumor is a leiomyoma.

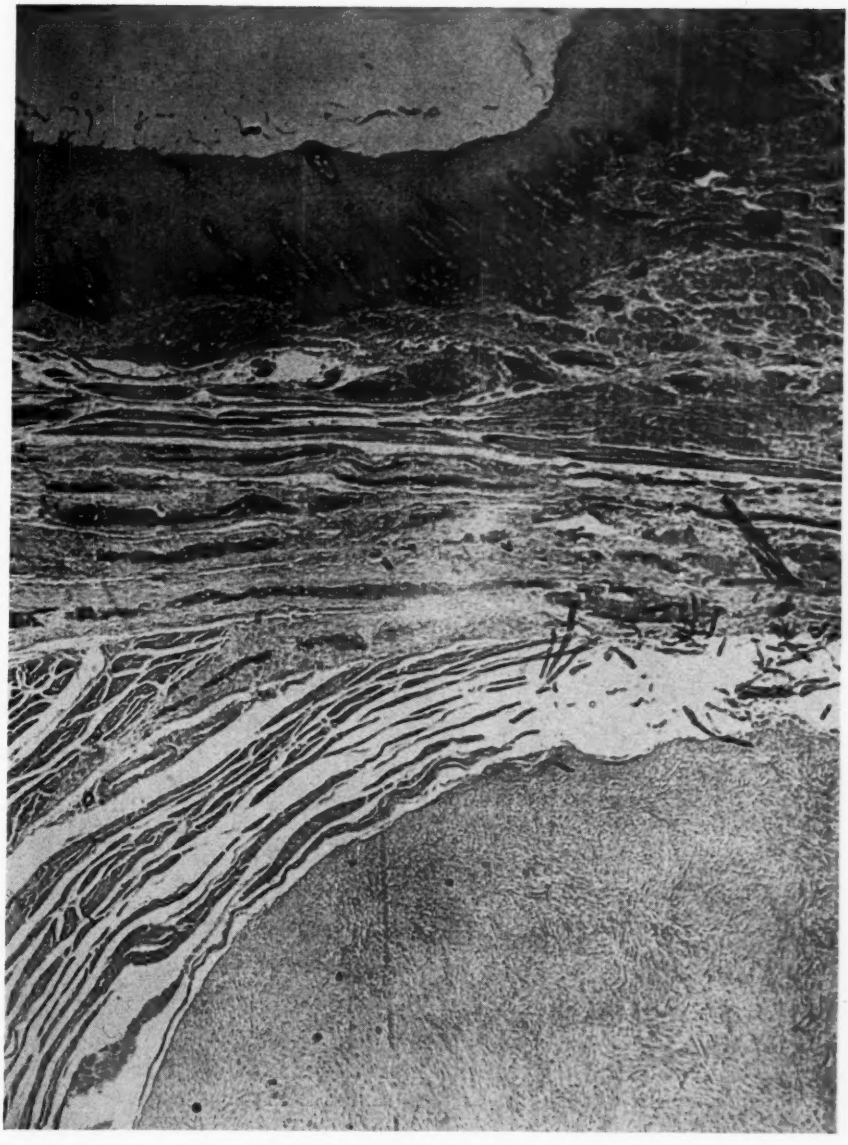

Fig. 3.—Magnified view of previous picture, showing independence of leiomyoma from the infiltrated carcinoma.

DISCUSSION

The etiology of both leiomyoma and carcinoma of the esophagus is largely unknown. The smooth muscle tumors are assumed to be congenital in origin, and some previously reported cases support this view. For example, Rose's case, where multiple leiomyomas occurred in the esophagus and stomach, could be interpreted as illustrating an anomaly confined to the primitive foregut. In

Fig. 4.— High-power view of leiomyoma (×200).

the myometrium, leiomyomas are common, and they can be produced experimentally by the use of estrogen. Rhythmical hormonal variation in concentration is believed to be the cause of the condition in human beings also, but there is no evidence to suggest that the esophagus is subjected to such humoral influences. Two of the factors thought to be predisposing causes in the pathogenesis of carcinoma of the esophagus, discussed by Wu and Loucks,[7] are illustrated in this case, namely: (1) a long period of consumption of hot liquids and foods and (2) a family history of cancer.

The duration of symptoms before the patient presented himself is one year. This is longer than the average in reported series of cases of carcinoma of the

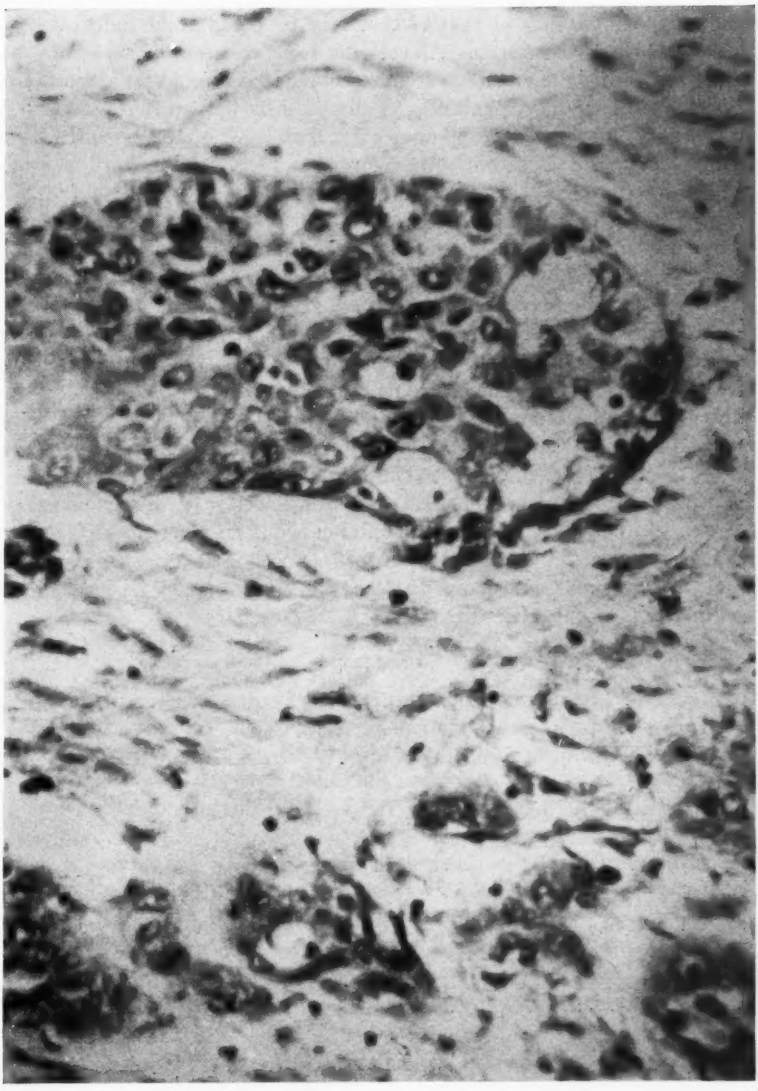

Fig. 5.—High-power view of carcinoma (×300).

esophagus. In Franklin and Shipman's[3] series of 159 cases, 60 per cent had symptoms for three months or less and only six patients had symptoms for a year or more.

It is tempting to suggest that the smooth muscle tumor must have played some active part. In the resected segment of the esophagus, the diameter of

the leiomyoma was 0.58 cm. The average width of the esophageal wall in these sections is 0.9 cm., so that at that level nearly 60 per cent of the thickness of the wall was due to the tumor. As sections from different parts of the block show the simple tumor to extend at least some distance around the circumference of the esophagus, encroachment on the lumen must have been present. The narrowing so produced was further accentuated by the superimposition of a carcinoma here. Fig. 2 shows these points clearly. In the barium swallow (Fig. 1), below the narrowed area, is a rounded shadow on the right, 1.25 cm. long, which could be a mucosal projection due to the underlying leiomyoma. Such an appearance is not characteristic of a malignant obstruction. It is recognized that points of constriction in the esophagus are more frequently the sites of carcinoma than are the intervening areas, according to Aird,[1] and it could, therefore, be argued that by accenuating this anatomical feature in a subject predisposed to carcinoma for other reasons, the simple tumor indirectly aided the development of the malignant one.

Previous investigators, including Rose,[6] held the opinion that solitary myomas are incidental findings and do not produce symptoms unless they are at least several centimeters in size. Yet, if a solitary tumor began to increase in size, it could produce the sequence of events described. In this instance, there is no evidence either way to form a firm opinion about the behavior of the myoma. On clinical grounds, the total length of symptoms is a feature suggesting that the leiomyoma was at first responsible for his symptoms, perhaps through recent growth in a concentric direction.

SUMMARY

1. A case of the simultaneous occurrence of squamous carcinoma and adjacent leiomyoma of the esophagus is described.

2. The relationship between the clinical picture and the pathology found is discussed.

This case was under the care of Mr. Donald Barlow, M.S., F.R.C.S., to whom I am indebted for permission to publish it.

Dr. K. W. F. Hinson did the pathological examinations and reported on the specimen. My thanks are also due to Mr. J. M. Webster for the photomicrographs.

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CHYLOTHORAX FOLLOWING A BLOW TO THE ABDOMEN

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ACCORDING to Zesas[1] the first description of a case of traumatic chylothorax was made by Bartolet in 1633. In 1946, Lampson[2] demonstrated that the condition could be cured by ligation of the thoracic duct in the lower chest. In 1938, Shackelford and Fisher[3] reported forty-one cases of traumatic chylothorax found in the literature, while Lampson found eighteen additional cases. Baldridge and Lewis[4] divided the cases into the following categories. Cases due to operative trauma were not included in Shackelford's series.

	SHACKELFORD AND FISHER 1938	LAMPSON 1947
Crushing injuries	17	1
Wounds (bullet and stab)	8	5
Fall from height	6	2
Blow on chest	5	2
Thrown against seat of automobile	4	--
Hyperextension	1	--
Auto accident with collision	--	2
Hit by automobile	--	1
Severe coughing	--	1
Surgical accident	--	4
	41	18

It is interesting to note that Shackelford and Fisher stated in 1938 that "surgical ligation of the duct is physiologically compatible with life, but clinically impractical at present." Since Peet and Campbell[5] reported the first successful ligation of the thoracic duct and Lampson reported the second successful ligation, a number have been done including our presented case which showed that ligation is clinically practical.

Traumatic chylothorax in children seems to occur rather frequently. Janet, Boegner, and Laquerrière[6] quote Kirchner, who reported the case of a child of 9 years of age who was pushed against a window bar by another child and fifteen days later developed a chylothorax. They also quote a case reported by Muttermilch of a 5-month-old infant who was dropped out of bed by its mother and in falling knocked against its cradle. It was apparently normal for two to three weeks and was then found to have a chylothorax. Sakula,[7] in 1949, stated that chylothorax in infancy is an extremely rare condition, and a search of the literature revealed thirteen cases under the age of 1 year, five of which occurred in the first four weeks of life. Sakula added another case in an neonate. In early infancy, usually designated as spontaneous after this age, practically all cases of chylothorax are associated with injury.

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CASE REPORT

S. C., a boy, aged 7 years, presented himself with symptoms of shortness of breath, especially upon exertion. The only clue was given by the patient who stated that three weeks previously he was hit in the "stomach" by a younger cousin and shortly after that became short of breath. Examination by palpation revealed nothing abnormal in the abdomen where the child seemed to center his attention, especially over and near the spleen. However, inspection of the chest revealed shallow respiration with some distention of the right side. Auscultation revealed normal breath sounds in the left lung but no breath sounds were heard on the entire right side. Percussion revealed dullness over the entire right chest. Fluoroscopically the left side was normal but the right side showed one solid shadow with no air visible. Pulse and temperature were normal; the patient weighed 56¼ pounds. He was admitted to St. Elizabeth Hospital.

An x-ray film taken of the chest on admission was read as follows: Films of the chest showed a rather massive effusion in the right pleural space which almost completely obscured the right lung. There was moderate displacement of the mediastinal structures to the left. Throughout the left lung fields there were multiple small rounded calcifications probably residual from tuberculosis or histoplasmosis. In view of these findings in the left lung it was possible that the effusion on the right side was tuberculous.

The same afternoon, July 29, 1953, under local anesthesia, 40 ounces of creamy fluid were removed from the right pleural cavity. The distention of the right chest seemed to be greatly diminished. The laboratory report of the fluid examination was as follows: Smear showed no organisms. There was no growth on culture. Total protein 15 Gm. per liter; sugar 93 mg. per cent; amylase 32 units; lipase 55 units; sp. gr. 1.012; total cholesterol 125 mg. per cent. Blood study showed the following: Hemoglobin, 12.5 Gm., red blood count, 4,230,000; white blood count 10,800; polymorphs, 58; lymphocytes, 40; monocytes, 2.

Films made of the chest the next day revealed that recheck following the thoracentesis showed a considerable decrease in the amount of the effusion in the right pleural space. There was, however, still considerable fluid remaining; the fluid extended up to the level of the fourth rib in the midaxillary line. Some air had been introduced into the pleural space and there was approximately 15 per cent collapse of the right upper lobe. The right upper lobe was not well visualized and showed diffuse miliary-like calcifications that were described in the left lung on previous study. The mediastinal structures were now close to the midline. Since the aspirated fluid was milky and since there was a history of previous trauma it was possible that the fluid accumulation could be chylous.

Clinically it could be determined that fluid was accumulating. A chest roentgenogram taken Aug. 5, 1953, revealed the left lung remained unchanged and a marked increase in the amount of fluid in the right pleural space. That same day the patient was tapped and 1,100 c.c. of milky fluid were obtained. Despite the fact that no more fluid could be obtained at the level of the seventh interspace posteriorly, films the next day showed very little change. During the stay in the hospital the patient's temperature remained close to normal while the respiration varied between 20 and 36. Due to administrative and financial difficulties he was dismissed from the hospital pending arrangements for future surgery.

He was readmitted on Aug. 17, 1953, and on that same day 1,400 c.c. of cream-colored fluid were withdrawn. On Aug. 21, 1953, 1,700 c.c. were withdrawn; on Aug. 26, 1953, 1,800 c.c. was withdrawn. An x-ray film made on Aug. 28, 1953, showed that the left lung remained clear while there appeared to be a slight decrease in the amount of fluid on the right. On Aug. 30, 1953, 2,000 c.c. of fluid were withdrawn and on Sept. 3, 1953, 1,200 c.c. were withdrawn. Recheck films made on Sept. 4, 1953, showed no apparent decrease in the amount of effusion since the previous x-ray study. There appeared to be some loculation of the fluid which was not evident on previous studies. There was still some displacement of the mediastinal structures. During this period the repeated aspirations kept the patient comfortable but did not succeed in keeping him dry.

On Sept. 5, 1953, the patient was operated on (M. R.). With the patient under satisfactory general anesthetic (Pentothal Sodium and nitrous oxide and ether), he was placed in

position for a right thoracotomy. The right pleural space was entered through the eighth intercostal space. Upon opening this, approximately 1,500 c.c. of chyle were removed by suction. The lung itself was markedly compressed. However, there were very few adhesions present and the pleural surfaces were glistening. The lung was reflected anteriorly and the posterior aspect in the region of the spine was examined. On the anterior surface of the lumbar spine, a small perforation of the mediastinal pleura was seen. Some lipomule was injected through the previously placed gastric tube and the fluid was again seen to flow through these perforations. Posterior to this the pleura was divided and the mediastinal pleura reflected anteriorly. Two intercostal vessels were divided between clamps just above the diaphragm. In doing this the aorta was reflected anteriorly and beneath this there was seen a ductlike structure which appeared to be the thoracic duct. At the region of the first lumbar vertebra there was some cystic dilatation. By blunt and sharp dissection the duct was dissected superiorly and found to be one structure. At this point it was divided between clamps and doubly ligated with silk. By gentle dissection the duct was dissected inferiorly at the diaphragm. At this point, however, it broke up into several branches. A right-angle clamp was placed across it at this point and the entire mass ligated en masse with double silk. The specimen was removed for study. There was some leakage of chyle, however, following this ligation; it was thought possible that the surrounding small collaterals had been injured. In order to obtain more reaction in the area some streptomycin powder was injected and some Gelfoam was placed at the site of amputation. The mediastinal pleura was then closed snugly with interrupted silk sutures. A drain was positioned laterally and placed under water drainage. The right lung completely expanded under positive pressure. The chest was closed in layers, using 0 chromic catgut throughout, with interrupted silk to close the skin. The patient withstood the procedure fairly well and returned to his room in good condition. One unit of blood and 500 c.c. of 1/6 molar lactate in 5 per cent glucose was used during the operation.

The biopsy reported dated Sept. 9, 1953 was as follows: "The specimen consists of three portions of tissue. One portion presents lobules of fat traversed by fibrous trabeculae and irregular channels. These channels appear to have valves and present an irregular scanty distribution of muscle fibers within their walls. There is also an incomplete cross section of a small artery and nerve trunk in this block of tissue. A second portion of tissue presents focally a peripheral desiccation and in relation to this focus there is an irregular accumulation of lymphocytes. The greater portion of the tissue consists of a collagenous matrix and small irregular lobules of fat. Throughout the matrix there are numerous irregular clefts and channels. These are of varying size. Within some of the channels there can be seen linings of cells; in others this is not definite. In the surrounding collagenous matrix there is a scanty irregular distribution of muscle fibers. In the two portions of submitted tissue the vessels seen are compatible with lymphatic vessels, but whether this represents an abnormality in structure I cannot say. There is no one large vessel but rather a plexiform arrangement of small vessels. Diagnosis: Vascular tissue."

Roentgenogram follow-up on the next day, Sept. 6. 1953, showed approximately 80 per cent collapse of the right lung with slight displacement of the mediastinal structures to the left indicating mild tension. There is little if any free fluid present. A recheck film on Sept. 7, 1953, showed almost complete re-expansion of the right lung and a small amount of free fluid present. On Sept. 9, 1953, an x-ray picture of the chest showed continuous good expansion of the right lung and the mediastinal structures in the midline. There was a slight increase in the amount of fluid in the right pleural space since the previous study. There were no hypostatic changes. On Sept. 15, 1953, x-ray films of the chest showed continued good expansion of the right lung. There was little if any fluid remaining in the right pleural space, but there was still some moderate pleural thickening.

It might be mentioned here that on Aug. 31, 1953, a total protein determination showed 4.9 Gm. per cent of which the serum albumin was 3.2 Gm. per cent and the serum globulin was 1.7 Gm. per cent. On Aug. 29, 1953, the report on the fluid removed on July 30, 1953, was negative for tuberculosis after pig inoculation. The patient made an uneventful recovery and was discharged on Sept. 17, 1953.

The patient was seen in the office on Dec. 11, 1953. He was feeling well. Weight was 58¾ pounds. Fluoroscopically, the entire right lung was fully expanded. The only abnormality seen was a thickening in the right lower area which somewhat obliterated the gutter.

CONCLUSION

Successful ligation of a ruptured thoracic duct was carried out with complete cure in a boy, aged 7 years, who was struck in the abdomen by a blow of a fist. A search of the literature failed to reveal another circumstance as this as the etiological factor. With the numerous opportunities for this type of trauma, especially in boxing, it is strongly suggested that a contracoup blow may have ruptured the thoracic duct at a congenitally weak or defective area.

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MITRAL COMMISSUROTOMY THROUGH THE RIGHT THORACIC APPROACH

Technique and Indications

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DURING the performance of surgery on the mitral valve in over 1,500 patients with rheumatic valvular heart disease,[1] several instances were encountered in which it would have been a distinct advantage to approach the mitral valve through the right hemithorax. This report is a description of a suitable technique and the indications for such an approach.

TECHNIQUE

A right thoracotomy is performed using either the posterolateral or anterior approach through the fourth intercostal space. After suitable exposure, the pericardium is opened anterior to the phrenic nerve. The mitral commissurotomy is carried out through the left atrial wall as follows:

The superior vena cava is further mobilized by dissection posteriorly in the groove between the cavae and the left atrial chamber (sulcus terminalis). After dissection, the most lateral portion of the left atrium is exposed. Two parallel rows of stay sutures are placed in this portion of the left atrial wall. An encircling purse-string suture circumscribes this entire area (Fig. 1).

Using the stay sutures for hemostasis, a stab wound is made through the left atrial wall, and the operator's ungloved left index finger tip is inserted for exploration of the left atrial chamber and the mitral valve. If possible, the mitral commissurotomy is then carried out by finger pressure; but if necessary, an instrument can safely be used in this position, although technically this procedure is more difficult than through the usual left auricular appendage approach (Fig. 2).

From this approach the relationship to the long axis of the mitral orifice is distinctly different from that of either the usual left anterolateral or posterolateral approach. Care in orientation is essential before attempting to open the valve.

After the mitral commissurotomy is completed, the opening in the atrial wall is approximated (by alternately crossing the previously placed stay sutures) and is then repaired with a continuous everting mattress suture of 000 silk or cotton swedged on an atraumatic curved needle.

Following the mitral valvular surgery, any additional procedure is completed in the manner necessary.

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INDICATIONS

I. Associated Valvular Lesions.—Among the patients with mitral stenosis, several have been encountered with an associated tricupid stenosis. While it is true that the right atrium can be explored through the left hemithorax (during performance of the mitral commissurotomy), the procedure is difficult; and, if the tricuspid valve is leathery and resists finger pressure, as is the usual case, it is almost impossible to perform the tricuspid commissurotomy safely with an instrument. (Although in a good many instances the mitral valve can be split by finger pressure alone, especially if calcified, the tricuspid valve rarely is calcified and in our experience, so far, has required cutting with an instrument in every instance.)

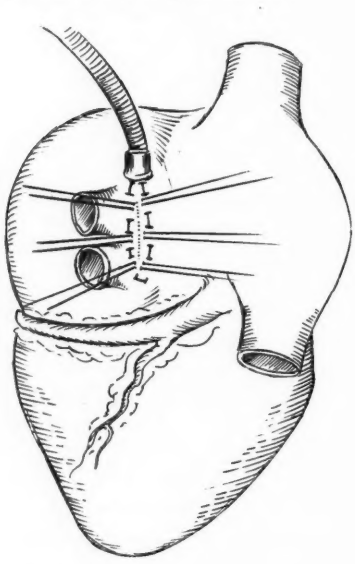


Fig. 1.

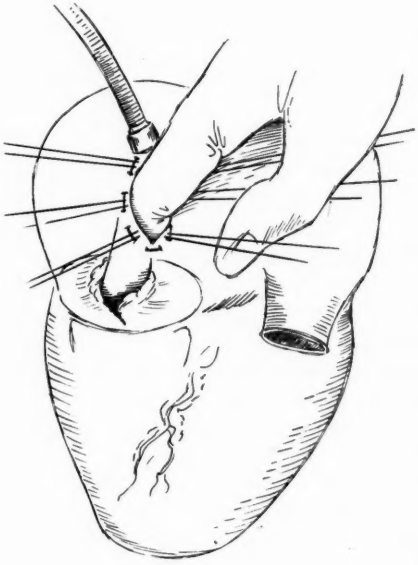


Fig. 2.

Fig. 1.—View of the right wall of the left atrium after dissection and placement of control sutures preparatory to exploration of the mitral valve.

Fig. 2.—View showing mitral commissurotomy being performed through the right lateral wall of the left atrium.

Before the present right thoracic approach was used, patients with mitral and tricuspid stenosis were first treated by mitral commissurotomy through the usual left thoracic incision. Then, at a later date, tricuspid commissurotomy was performed through the right thoracic approach.[2] This not only required two operations, both of which were performed on a very ill patient, but also necessitated bilateral thoracotomy. At the present time we are performing both operations simultaneously on such patients through an incision in the right hemithorax. The tricuspid commissurotomy is done through the right auricular appendage (Fig. 3), and then the mitral commissurotomy is performed by the method described.

II. Associated Pulmonary Pathology.—In the past we have encountered several patients with associated pulmonary pathology of various types. Several patients with bronchiectasis, localized to the left side, have had combined mitral commissurotomy and pulmonary resection. Although we have performed mitral surgery on patients with bronchiectasis localized to the right side, they have caused us the expected postoperative trouble due to increased secretions in spite of intensive medical management.

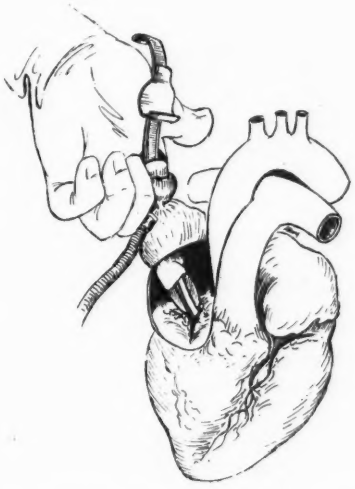


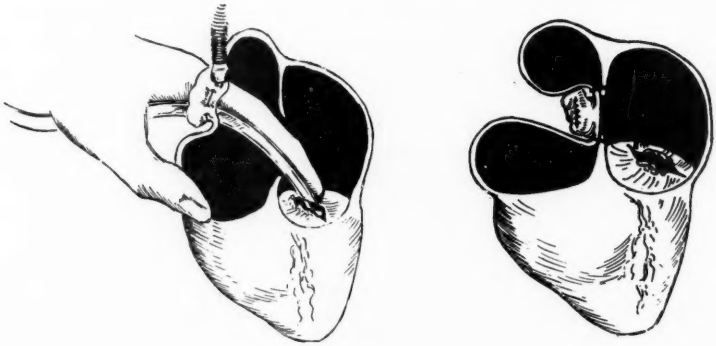
Fig. 3.—Technique of tricuspid commissurotomy.

In the future (when surgical treatment is deemed necessary) in patients needing mitral valvular surgery and having bronchiectasis localized to the right side, the approach through the right hemithorax will be used. If both procedures are performed, the patient not only should escape a second operation, but also should have an easier postoperative course.

Recently we had a patient with mitral stenosis and an obscure shadow in the right lung. All preliminary studies were negative with regard to the pulmonary pathology, and since the mitral stenosis was not producing marked symptoms at the time, the right chest was explored first and an infareted right middle lobe was found. This lobe was resected, and postoperatively the patient had an extremely troublesome course due to cardiac complications. The patient finally recovered and returned three months later for an uneventful mitral commissurotomy.

With the present approach we have operated on an elderly, extremely ill patient with a somewhat similar roentgenographic lesion in the right lung. This patient proved to have a bronchiogenic carcinoma, in the right upper lobe; although quite ill from a cardiologic viewpoint, he withstood, quite well, simultaneous mitral commissurotomy and resection of the right upper lobe together with a block resection of the chest wall (Case 1).

III. Associated Cardiac Anomalies.—Patients with Lutembacher's syndrome are ideal for such an approach. It is an easy matter to explore the mitral valve through the interatrial septal defect, and after commissurotomy the defect is repaired by the technique of atrio-septo-pexy which has previously been recorded (Fig. 4).[3, 4]



A. *B.*

Fig. 4.—*A*, Mitral commissurotomy being performed through the interatrial septal defect. *B*, Closure of the septal defect by the technique of atrio-septo-pexy.

CASE REPORTS

CASE 1.—H. S. (H.H. 90968), a 55-year-old white man, entered Hahnemann Hospital on Aug. 20, 1953, complaining of three-pillow orthopnea of three months' duration, following, supposedly, a right pulmonary infarction.

Rheumatic fever had been diagnosed at the age of 13 years, and cardiac insufficiency had occurred three years before admission, the chief complaint had been dyspnea on exertion. This was fairly well controlled with digitalis. One year before admission he had suffered a left cerebral embolization with a repetition six months later on the right. Functional recovery from both had been excellent.

Physical examination revealed an orthopneic man with a pulse of 80, and blood pressure of 120 systolic and 90 diastolic. The cardiac rhythm was that of auricular fibrillation. The mitral first sound was sharp and accentuated. There was a midlate, rumbling diastolic murmur with presystolic accentuation, best heard in the mitral area. The second pulmonic sound was accentuated.

Fluoroscopy and roentgen examination revealed an area of increased density in the posterior upper portion of the right lung field. The cardiac configuration revealed two plus enlargement of the right ventricle and left atrium (Fig. 5). There was also evidence of calcification of the mitral valve. The electrocardiogram revaled auricular fibrillation, right ventricular hypertrophy, and digitalis effect. Bronchoscopy showed no evidence of endobronchial disease; examination of bronchial secretions for tumor cells was negative.

The preoperative impression was rheumatic valvular heart disease with mitral stenosis and an undiagnosed parenchymal lesion in the right lung.

On Aug. 27, 1953, an exploratory operation was performed through a right lateral thoracic approach. The right lung was examined and a tumor was found to be present in the upper lobe with extension into the chest wall. The right upper lobe was resected together with a block of chest wall containing the third, fourth, and fifth ribs. Mitral commissurotomy was then carried out by the technique described. The mitral valve was found

to be calcified and had an opening about 5 mm. in diameter. The valve was split by finger pressure to an opening of 25 mm. in diameter without the production of any regurgitation.

The patient withstood surgery quite well. Pathologic examination of the resected right upper lobe revealed squamous cell carcinoma, Grade III. He was discharged on Sept. 12, 1953 (sixteen days postoperatively), at which time he was considered to be in good general condition (Fig. 6).

CASE 2.—F. F. (H.H. 91086), a 39-year-old white woman, entered the hospital on Aug. 24, 1953, complaining of dyspnea and fatigability of ten years' duration and anasarca for three years' duration. Abdominal paracentesis every two months had been necessary for the past year. Rheumatic fever had been diagnosed at the age of 17 years. She had been on a low salt diet, mercurial diuretics, and digitalis.

Physical examination revealed a blood pressure of 120 systolic and 70 diastolic. Auricular fibrillation was present. At the mitral area there were heard a sharp first sound and a midlate diastolic rumble with presystolic accentuation. In the tricuspid area, when the patient was placed in the right lateral position, there was a diastolic murmur which appeared to be different from that heard in the mitral area. The lung fields were clear. The liver was enlarged 5 cm. below the costal margin. Pitting edema of the lower extremities and evidence of abdominal fluid were also present.

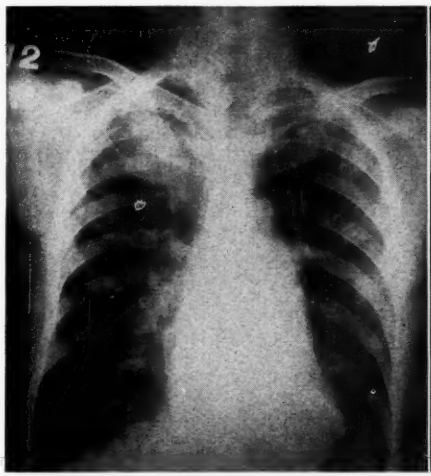


Fig. 5.

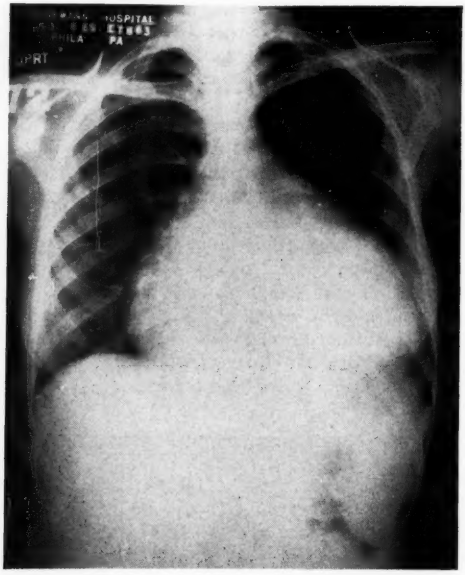
Fig. 6.

Fig. 5.—Preoperative roentgen view demonstrating the parenchymal lesion in the right upper lung field. There is also the typical cardiac configuration of mitral stenosis.

Fig. 6.—Postoperative view following simultaneous mitral commissurotomy and resection of the right upper lobe together with a portion of the chest wall.

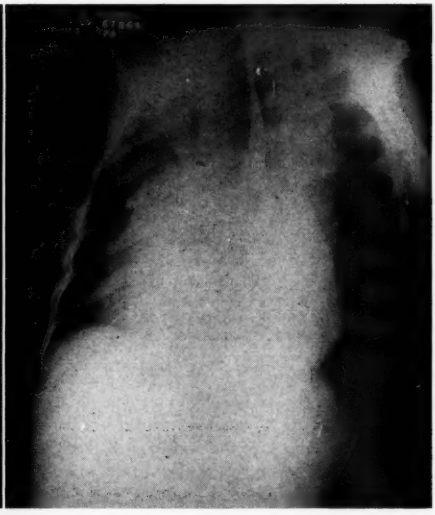
Fluoroscopy and roentgen examination showed an enlarged right ventricle and left atrium, and questionable enlargement of the right atrium (Fig. 7, *A*, *B*, and *C*). An electrocardiogram revealed right ventricular hypertrophy and auricular fibrillation. The remaining laboratory examinations were normal. On the basis of the clinical findings it was the preoperative opinion that this patient had combined tricuspid and mitral stenosis. In view of the presence of auricular fibrillation it was not deemed of value to perform cardiac catheterization for physiologic evaluation of the tricuspid valve.

On Aug. 31, 1953, an exploratory operation was performed through the right thoracic approach. The right atrium was found to be tremendously enlarged and the opening of the tricuspid valve was less than 10 mm. in its longest diameter. The valve was quite



A.

B.



C.

Fig. 7.—*A*, Preoperative view showing cardiac enlargement. *B*, Demonstration of enlargement of the left atrium and right ventricular outflow tract. *C*, View showing enlargement of the right atrium.

leathery and resisted commissurotomy by finger pressure alone, so the valve was cut with the guillotine knife until an opening of about 25 mm. was obtained. Following tricuspid commissurotomy, the mitral valve was explored by dissecting the sulcus terminalis and passing the finger through the right wall of the left atrium. Its opening was found to be about 4 mm. in diameter. By means of commissurotomy an opening of about 18 to 20 mm. was obtained without regurgitation.

Postoperatively, the patient had a relatively uneventful hospital course. She was discharged on Sept. 29, 1953, twenty-nine days following surgery (Fig. 8).

CASE 3.—J. R.[4] (Samaritan Hospital, São Paulo, Brazil), a 31-year-old white man, entered the hospital with marked progressive physical incapacity and evidence of congestive heart failure of two years' duration.

Physical examination revealed a sharp mitral first sound and a late diastolic rumble with presystolic accentuation. A systolic murmur was present over the entire precordium.

Roentgen and fluoroscopic examination of the heart revealed marked cardiac enlargement. Preoperative cardiac catheterization studies demonstrated a left-to-right shunt of 6 L. per minute at the atrial level. It was the preoperative opinion that the patient undoubtedly had Lutembacher's syndrome.

On Feb. 13, 1953, the patient was explored through the right thorax. Digital exploration of the right atrium revealed a centrally located defect in the interatrial septum, about 3 cm. in diameter. Examination of the mitral valve, through the septal defect, disclosed a very severe mitral stenosis. Mitral commissurotomy was performed by finger pressure through the septal defect. Atrio-septo-pexy was then accomplished in the manner previously reported. The patient stood the procedure very well.

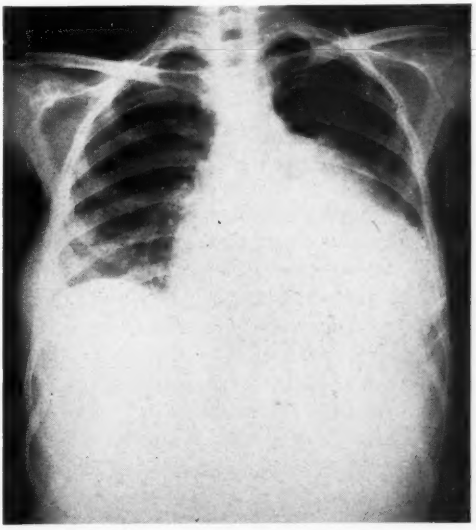


Fig. 8.—Postoperative view following mitral and tricuspid commissurotomy through the right thoracic approach.

SUMMARY

1. A suitable technique for performing mitral commissurotomy through a right thoracic approach is described. The procedure is safe and practical.

2. The indications are outlined. By the use of such an approach, patients with mitral stenosis and associated lesions such as tricuspid stenosis, inter-

atrial septal defects, and pulmonary disease in the right lung can be treated completely at one operation. This offers the obvious advantage to the patient of a single operation, and also, in many instances, actually increases the chance of survival by decreasing the chance of morbid complications.

3. Three cases are reported in detail to demonstrate the indications and the desirability of such an approach.

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UNUSUAL PROBLEMS IN MANAGEMENT OF CHEST WALL DEFECTS

Report of Two Cases

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DEFECTS of the chest wall most commonly follow replacement of normal structures by neoplastic tissue which in turn requires wide surgical excision. Less frequently, radiation necrosis produces extensive tissue loss. Much less frequently, trauma, followed by surgical treatment and complicated by infection, may produce defects of major importance. According to Effler and Blades,[12] the low incidence of traumatic defects during World War II may be attributed to splendid early surgical therapy together with the use of antibiotics. These authors further state that, in most instances, the patient's regional tissues may be employed satisfactorily for closure without need of foreign material of various kinds.

Repair of defects of the superficial soft tissues of the chest wall does not involve surgical principles different from those followed in other parts of the body. However, when the bony structures of the cage are destroyed, problems peculiar to the repair of thoracic defects arise. In this article we have purposely omitted discussion of chest wall defects associated with existing bronchopleurocutaneous fistula since these latter situations demand additional attention to the underlying problem of origin and ablation of the fistula.

Lung hernias have been cured by simple pressure bandages[27] or by packing with irritant-soaked gauze.[15] As long ago as the end of the last century Tuffier[35, 36] repaired a lung hernia by simple ligation of the sac. Volger[38] later suggested the use of osteoperiosteal flaps which subsequently have been used by many other surgeons in the repair of chest wall defects.[1, 14, 18, 25, 27, 30]

Free rib segments have been used to stabilize the chest wall after operative correction of pectus excavatum. Dailey[9] was first to employ this method, which was subsequently used by others.[2, 10] Kirschner wires have been used for the same purpose.[26] Free rib segments have rarely been employed to stabilize the chest wall following other procedures.[11] Winkel,[40] in 1935, used woven fascia lata strips to anchor defect margins, while Watson and James[39] used large plaques of the same fascia anchored in place by interrupted sutures. Defects created by drainage of empyema cavities have been successfully lined by pedicle grafts.[4, 28]

Campbell[8] reported a clever method for reconstruction of anterior chest wall defects by using the latissimus dorsi muscle. As a prerequisite for success of this method, the subscapular vessels must be intact. The pectoralis major muscle has also been utilized to bridge anterior wall defects.[23] The mobilized female breast was first used to close a chest wall defect by Sauerbruch.[33] Re-

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cently Maier,[24] Pickrell and his associates,[31, 32] and others[20] have had gratifying success with this procedure. Pickrell[31, 32] reported the use of a free split thickness graft applied directly upon the anterior pericardium after resection of the lower sternum and costal cage. Urban[37] and Pickrell[31, 32] have recorded nice results using adjacent thoracic and abdominal soft tissue flaps. One of us (T. D. C.) has successfully employed such flaps to repair defects of the anterior chest wall following excision of radionecrotic and malignant tissue, the result of breast disease.

Tantalum plates have been employed in rare instances but without striking success.[3, 7, 16, 30] In general, the plate method has been abandoned.

Tantalum and/or stainless steel mesh gauze have been used with real satisfaction by numbers of surgeons.[1, 11, 20, 29] In general, of the foreign materials employed, the metallic mesh has been most satisfactory.

The reports here presented concern two patients in each of whom trauma with superimposed infection, followed by surgical therapy, resulted in a large incapacitating defect of the chest wall.

CASE REPORTS

CASE 1.—E. A., a 38-year-old white man, was admitted to Veterans Administration Hospital, Houston, Texas, on Sept. 14, 1949, with a history of having received a penetrating, small caliber missile wound of the left chest while in combat in 1944. The left chest was explored and drained, soon after the injury. Postoperatively the patient developed an empyema manifest by drainage of purulent exudate from the left chest. Because of persistence of drainage, together with hemoptysis, a partial thoracoplasty was done. Following this procedure he did well with the exception of repeated small episodes of hemoptysis, weakness, and dyspnea. In April, 1949, he experienced a severe pulmonary hemorrhage. The presenting complaint was a small episode of pulmonary bleeding three days before admission.

Physical Examination.—The patient was a poorly nourished white man who appeared chronically ill. Vital signs were normal. There was marked deformity of the left chest with partial absence of the anterior or lateral aspects of ribs, two through nine inclusive. The old thoracoplasty scars and gunshot wounds were well healed. There was marked limitation of motion of the left chest with dullness on percussion. Bronchial breath sounds with crackling râles were heard over the left lung. The remainder of the physical examination was noncontributory.

Accessory Clinical Findings.—Laboratory findings were all within normal limits. These included repeated negative sputum studies for tubercle bacilli.

Course in the Hospital.—Chest roentgenograms at the time of admission revealed an absence with partial regeneration of the left second, third, and fourth ribs anteriorly and the fifth, sixth, seventh, eighth, and ninth ribs laterally. Clear lung areas were noted in the apical portion on the left with a slight shift of the heart and mediastinum to the right. X-ray appearance suggested that a partial pulmonary resection had been done followed by thoracoplasty. Bronchographic examination, on the other hand, showed filling of the entire left lung with marked compression of the basilar segments (Fig. 1). Bronchoscopic examination revealed active bleeding from the left upper lobe.

On Oct. 24, 1949, an exploration of the left hemithorax revealed a dense, scarred, spherical mass in the lower portion of the left upper lobe which contained a foreign body, thought to be a piece of camouflage net. Small loculations of purulent exudate were encountered. The left lung was collapsed and nonaerated. A pneumonectomy was performed. Postoperatively the patient developed a spiking temperature and evidence of empyema. Purulent exudate was aspirated from which *Staphylococcus aureus* was cultured. The patient was treated with antibiotics, repeated irrigation of the chest cavity, and finally by open drainage. The general

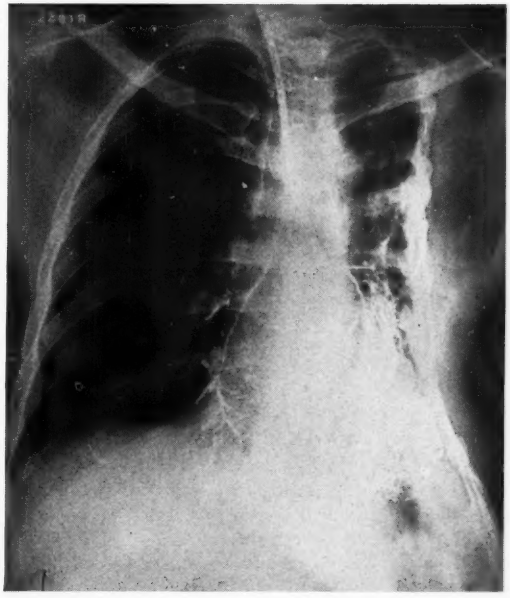

Fig. 1 (Case 1).—A bronchogram done soon after admission revealed the presence of both lobes on the left with diminution in size of the left pleural space.

Fig. 2 (Case 1).—The aorta and left side of the heart are clearly seen in this late photograph taken over one year after completion of chest wall surgery.

condition, thereafter, improved. Several focal revisions of the thoracoplasty wound were done; the space was irrigated and packed. These operations left an unroofed, narrow, deep cleft in the left hemithorax, bounded medially by the mediastinum and vertebral bodies and laterally by the subscapular tissue and regenerated rib segments. In spite of these procedures, it appeared that the defect would not heal by secondary intention and therefore, on Sept. 7, 1950, a split thickness skin graft was applied within the graunulation tissue-lined defect. The split thickness skin graft took well. Later minor revisions included excision of the scapula tip before complete, permanent wound closure was obtained (Fig. 2).

CASE 2.—J. A. M., a 49-year-old white man, was admitted to the Veterans Administration Hospital, Houston, Texas, on March 24, 1951, with complaints of chronic cough and dyspnea. In 1918, while in combat in France, he received a gunshot wound of the right chest. He was discharged after nine months' hospitalization, but continued to have a chronic cough. In 1928, ten years after his injury, a right lung abscess was demonstrated. This was treated by repeated aspirations but he remained chronically ill with frequent febrile episodes. In 1947, a right pneumonectomy was performed at a Navy hospital. On the fifth postoperative day, there was a blowout of the bronchial stump. This was treated by closed thoracotomy drainage for nine months. He was transferred to the Veterans Hospital, McKinney, Texas, weighing 87 pounds; in February, 1948, a Schede type thoracoplasty with closure of the bronchial stump was done. Following this procedure, drainage stopped but the patient had marked respiratory distress; this was complicated by asthma.

Physical examination on arrival at our hospital revealed a chronically ill man, moderately dyspneic even at bed rest. There was marked asymmetry of the thorax. There was an old scar from the previous thoracoplasty, which extended from the superior border of the scapula posteriorly to the sixth rib anteriorly; there was marked depression of the right side of the chest. Movements of the chest wall on respiration were paradoxical. On the right side, the heart was covered only by skin, subcutaneous tissue and pericardium and the pulsations were quite prominent. No ribs could be palpated laterally above the level of the tenth. On inspiration there was a shift of the lower ribs medially (Fig. 3). The remainder of the physical examination was noncontributory.

Accessory Clinical Findings.—Chest roentgenograms revealed evidence of an extensive thoracoplasty on the right with minimal bony rib regeneration (Fig. 4). There were no lung markings on the right side. The left lung was clear and, indeed, perhaps overaerated. Fluoroscopy revealed considerable mobility of the mediastinum. There was marked irregularity in the descent of the diaphragm on the right side with respiratory excursions. In addition, inefficient use of the left diaphragm was also noted. With external support of the right chest, better function of the left diaphragm and better stabilization of the mediastinum were noted. Respiratory studies were made and revealed marked impairment of function. copy revealed considerable mobility of the mediastinum. There was marked irregularity in Because of the paradoxical movements of the chest wall, which had not been reduced materially by corsets, an attempt at stabilization seemed desirable. This was done on July 13, 1951.

The old scar was excised and skin and subcutaneous tissue flaps were freed from the subjacent pericardium. The fourth costal cartilage was excised exposing the border of the sternum; the body of the eighth thoracic vertebra was exposed posteriorly.

The eleventh and twelfth ribs were then removed through an accessory oblique incision, following which these ribs were inserted as bony struts. The eleventh rib was attached posteriorly to the eighth vertebral body with two screws; its anterior end was attached with stainless steel wire to the lateral edge of the sternum at the site of removal of the fourth costal cartilage. The twelfth rib was then bridged between the middle of the newly transplanted eleventh rib and the undisturbed tenth rib, thus forming a "Y." The attachment between ribs was accomplished with stainless steel wire. The wound was then closed in layers with a drain beneath the flap. A pressure dressing was applied. Postoperatively, the patient did well and the dyspnea was greatly improved (Fig. 5).

Fig. 3 (Case 2).—The depression of the chest wall on inspiration is readily apparent.

Fig. 4 (Case 2).—This film illustrates the status of the bony chest wall at the time of admission.

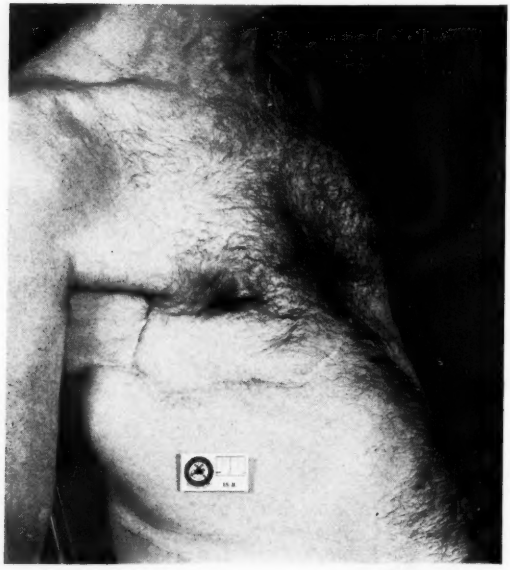

Fig. 5 (Case 2).—This photograph, also taken during inspiration, was made several months postoperatively. Note the absence of the marked depression which existed before operation.

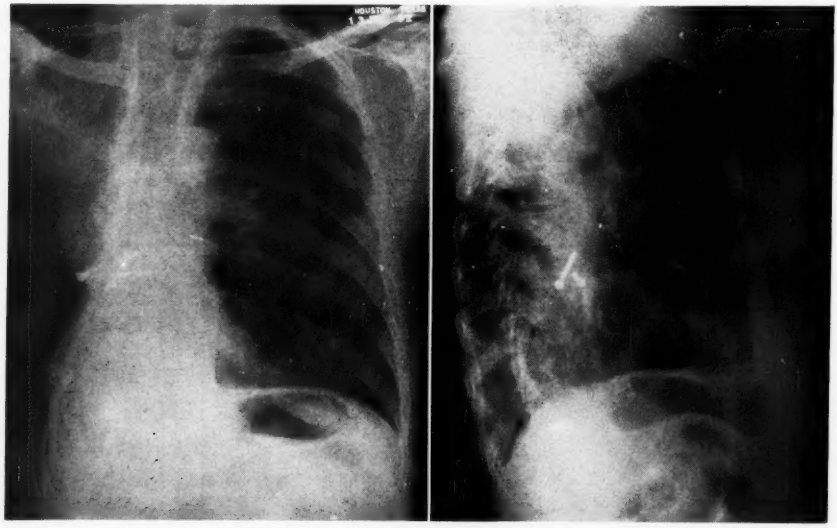

Fig. 6 (Case 2).—These films show the rib struts in place. Similar films made a year later show no change.

He was last seen on Sept. 16, 1953, two years following operation, at which time he was cheerful and stated he was much better than before the reconstructive procedure. He was free of dyspnea at rest and even with slight activity, but he has little respiratory reserve and becomes dyspneic on moderate exertion. The rib grafts are still in place, as shown by palpation and x-ray films (Fig. 6). He has slight motion just below the rib grafts, and there is still some medial displacement of the lower cage on inspiration.

DISCUSSION

In Case 1, a difficult problem was finally solved in a relatively simple fashion. After attempts to obliterate the space by collapse measures had failed, the space was marsupialized. No previous mention of the use of split skin grafts for this purpose has been found, though pedicle flaps have been employed. The method has the disadvantage of leaving an unsightly deformity.

The problem involved in the second case was paradoxical motion of the chest wall. This sometimes becomes a factor of great importance when a large section of the rib cage is removed. In our review of the literature, we found only rare instances in which free rib grafts were employed in reconstruction of the chest wall. While the use of autogenous material seems desirable, it is reasonable to suppose that bank bone might be used to advantage for this purpose.

SUMMARY

The recent literature on management of chest wall defects has been reviewed. Two illustrative case reports have been presented.

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PRESSURE BREATHING IN SURGERY

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PRESSURE breathing is the control of respiratory dynamics by the deliberate alteration of intrapulmonary pressure. It is most commonly produced by the application of pressure by means of a mask or an endotracheal tube. Through pressure variations one can control respiratory exchange, facilitate surgical procedures, or, under certain circumstances, stabilize circulatory function.

Pressure breathing is used in artificial respiration to improve ventilation. It is employed during general anesthesia where powerful relaxant drugs have been used to facilitate surgery. Here the anesthetist may attempt to maintain satisfactory ventilation by aiding the inspiratory effort of the patient. This is termed "assisted respiration"; or the anesthetist may render the patient apneic and assume responsibility for the rate as well as the volume of ventilation. This is known as "controlled respiration." The open chest, in the majority of instances, adversely affects respiratory dynamics. Pressure breathing is, therefore, also indicated in this situation.

There seems to be very little criticism of the need for pressure breathing. There is, however, much that can be said against its application, for, if it is improperly applied, the patient may be injured.

Although pressure breathing had long been employed for resuscitation, it was not until the advent of surgical entrance into the thoracic cage that it was employed in surgery. Surgeons have played an important part, not only in recognizing the need for pressure breathing, but also in the development of pressure-breathing apparatus. Mikulicz, in 1903, recognizing that intrathoracic surgery could not progress until the hazards of an open thorax could be overcome, commissioned his student, Sauerbruch, to resolve the problem. Sauerbruch,[11] recognizing the need for some form of pressure breathing, built, and in the following year presented, the pressure chamber known by his name (Fig. 1). The patient's body is within the negative pressure cabinet. The patient's head extends into the surrounding room. This diagram of the Sauerbruch cabinet is extremely interesting. It reveals, perhaps, in some new manner, the brilliance of Sauerbruch. As you may note, the lower part of the patient's body is in communication with the outside room and is thus maintained at atmospheric pressure. One must therefore assume that Sauerbruch was familiar with something that we have but recently begun to appreciate, that is, that in our endeavor to prevent the

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hazards of pneumothorax by pressure breathing, we introduce new dangers. It is also evident that he appreciated the desirability of not interfering with venous return to the right heart under these circumstances.

In 1908 Meyer[8] presented his double-chamber apparatus (Fig. 2). The patient's body is maintained under negative pressure and the patient's head is in a positive pressure cabinet. It seems clear that Meyer either did not believe, or failed to appreciate, some of the deleterious effects of pressure breathing which were of such vital concern to Sauerbruch. It is perhaps of particular interest to this group that Murphy[9] and Robinson,[10] and associates,[10] all in Boston, contributed to the literature on pressure breathing by the creation and description of new apparatus.

After the early years the progress in pressure breathing rested to a large extent in the hands of the anesthesiologist. An important advance was the development of controlled respiration, that is, the production of apnea and ventilation by manual bag compression. A surgeon, Crafoord,[1] devised a mechanical means for controlled respiration, and, more recently, Maloney and associates[4-7] have contributed much to this important subject.

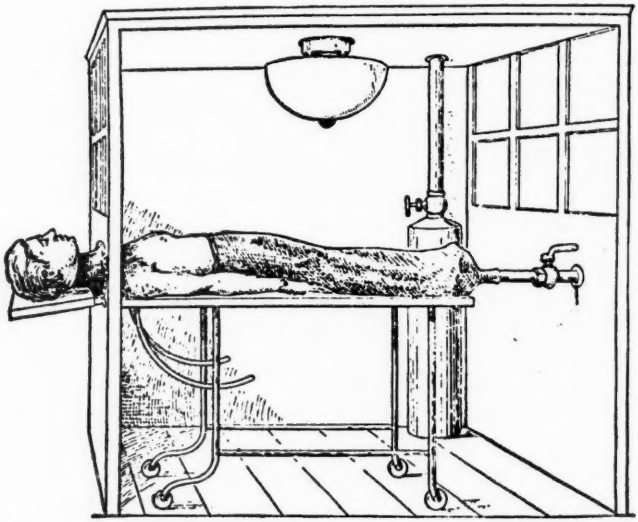

Fig. 1.—Sauerbruch cabinet. Position of patient in chamber ready for operation under negative pressure.

It is perhaps important to review the possible effects of increased intrapulmonary pressure on a physiologic basis. In the seventeenth century Valsalva recognized that increased intrapulmonary pressure interfered with the flow of blood through the lungs. He determined that ". . . if, after a deep inspiration, a strenuous and prolonged expiratory effort be made against a closed glottis, the pressure so exerted on the heart and greater vessels, arrests the flow of blood through the lungs." That such increased intra-

pulmonary pressure can cause a fall in arterial pressure was noted by Einbrodt.[3] He stated that positive pressure breathing produces a great fall in arterial pressure due to "the inability of blood to enter the thoracic cavity, so that the heart becomes depleted and its output falls." Weber,[12] stated, "I discovered . . . I could interrupt at will for a moment, the beats of my heart and the pulsation of my arteries by hindering the air from entering into my chest which I was at the same time compressing."

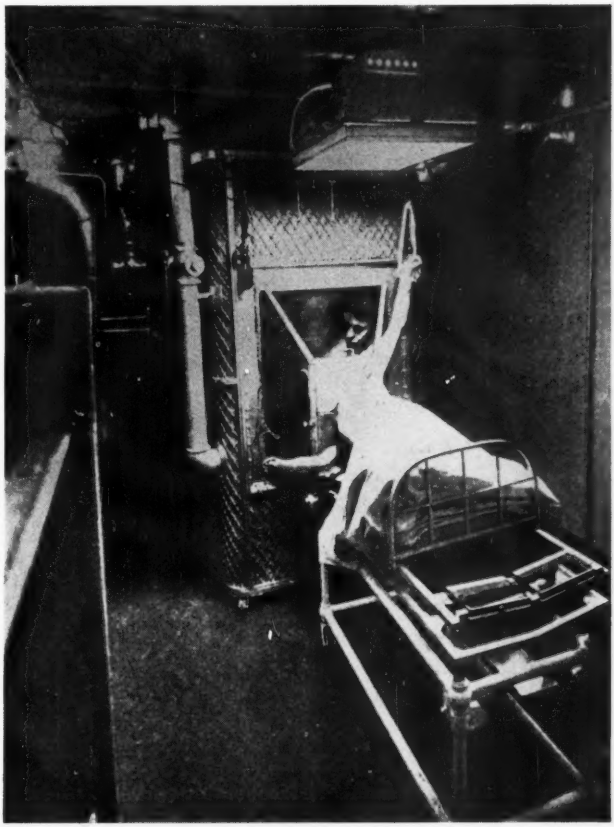

Fig. 2.—Willy Meyer double-chamber apparatus.

More recently, many have shown that positive pressure breathing results in a diminution in cardiac output. The mechanism seems to be clear (Fig. 3). We note from the right side of this drawing that increased intrapulmonary pressure produces an increased vascular resistance because of direct pressure upon the capillaries within the lungs. Interference with the perfusion of blood across the capillaries of the lung results in a decreased left ventricular filling. A decrease in left ventricular filling is followed by a decrease in

cardiac output. On the left portion of the drawing we note that increased intrapulmonary pressure causes cardiac tamponade. Tamponade results in poor filling of the right heart; this in turn means a decreased perfusion of blood across the pulmonary capillary bed, followed in turn by a decrease in left ventricular filling and a consequent decrease in cardiac output. It is important to note that since right atrial pressure is increased, there is a backing up of venous blood with an elevation of venous pressure.

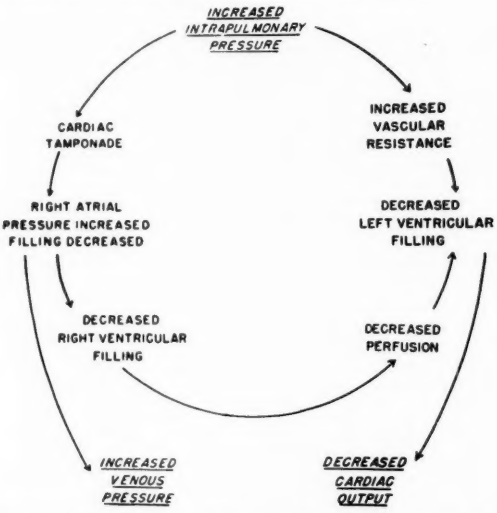

Fig. 3.—Effect of increased intrapulmonary pressure on cardiac output and venous pressure.

The patient in good condition can tolerate a moderate amount of increased intrapulmonary pressure. The patient in a poor physical state cannot. The ability to withstand positive pressure breathing depends to a great extent on the patient's ability to maintain a heightened venous pressure, for, during the phasic drop in intrapulmonary pressure, the elevated venous pressure is an important factor in filling the right heart. It thus contributes to the maintenance of a satisfactory cardiac output. Nothing should be done which will interfere with the patient's ability to develop and maintain an elevated venous pressure.

Ganglionic blockade effectively prevents the patient from elevating his venous pressure. Thus, ganglionic blockade, as for hypotensive anesthesia, or by spinal or paravertebral block, renders these patients poor candidates for pressure breathing. It has been established beyond doubt that hypovolemic patients tolerate pressure breathing extremely poorly. These patients are incapable of maintaining a satisfactory venous pressure, let alone being able to elevate it. Blood loss in a patient under pressure breathing is a hazard out of all proportion to the amount of blood lost, and in these patients particularly, blood replacement is vital and should not be delayed. It should be anticipated.

Since pressure breathing is an important aid to some forms of abdominal surgery, and a vital adjunct to intrathoracic surgery, means to decrease its hazard should be employed.

The effect of pressure breathing on cardiac output seems to be in direct ratio to the mean pressure. The greater the mean pressure, the greater is the effect on cardiac output. The mean pressure can be lowered by intermittently permitting the intrapulmonary pressure to drop to ambient pressure. The

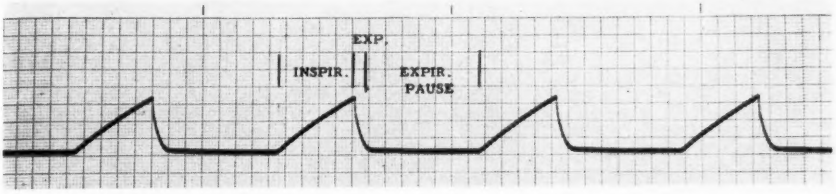

Fig. 4.—Phases of inspiratory positive pressure respiration.

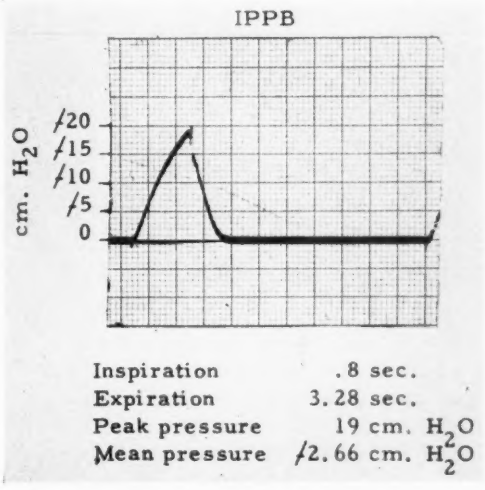

Fig. 5.—Inspiratory positive pressure respiration with low mean pressure.

duration of expiration should be longer than the period of inspiration, to allow the heart to catch up during this period of phasic decrease in intrapulmonary pressure (Fig. 4). This latter is more easily accomplished in controlled than in assisted respiration.

A protracted period of elevated intrapulmonary pressure produces deleterious effects on circulation and gas exchange. It is the practice of many thoracic surgeons to request such a period of heightened intrapulmonary pressure. Request is made of the anesthetist during the closure of the chest to bring the lung to the chest wall and keep it there, through the period of such closure. Of all the circumstances, and of all the times that a high mean

Fig. 6.

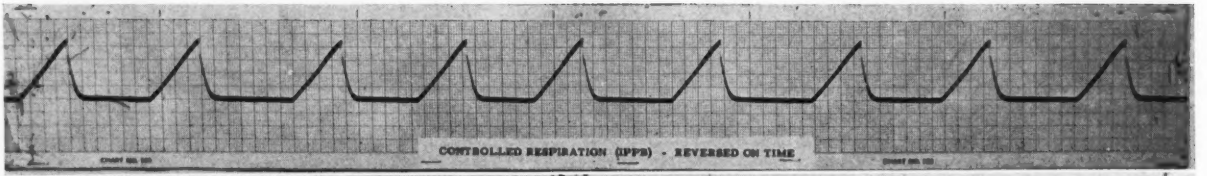

Fig. 7.

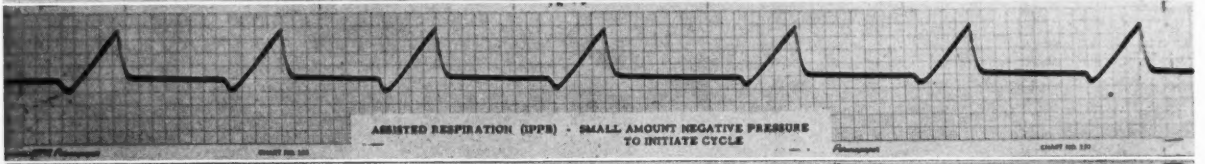

Fig. 8.

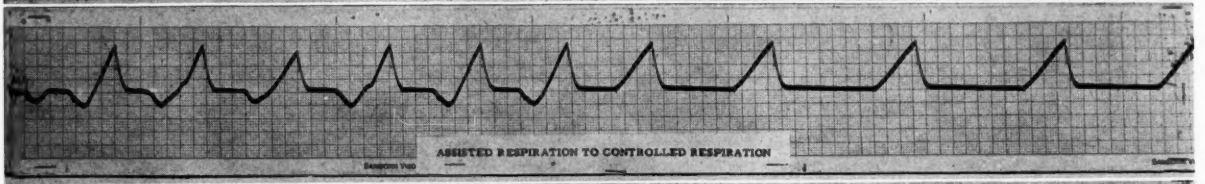

Fig. 9.

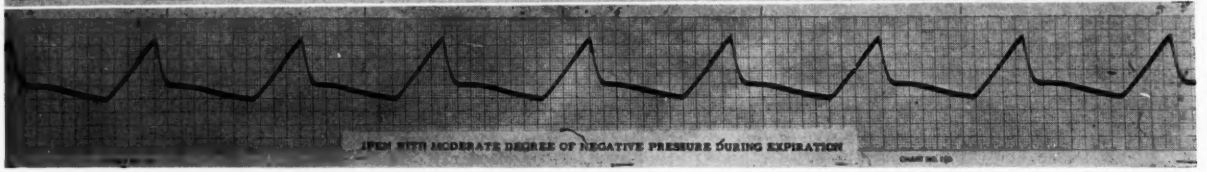

Fig. 6.—Controlled respiration, inspiratory positive pressure breathing.
Fig. 7.—Assisted respiration, inspiratory positive pressure breathing.
Fig. 8.—Assisted to controlled respiration.
Fig. 9.—Controlled respiration, inspiratory positive-expiratory negative pressure breathing.

intrapulmonary pressure may be particularly harmful, none exceeds this set of conditions. It is especially at this time that nothing should be done further to compromise the patient. It is our experience that chest walls can be safely closed and pneumothorax be prevented by intermittent insufflation of the lung and by chest aspiration.

During thoracic surgery occasional inflation of the atelectatic lung is desirable. The collapsed portion of the lung should be inflated carefully by intermittent positive pressure breathing with increasing increments of pressure at the peak of inspiration. Rapid inflation of the lung by constant positive pressure is undesirable and harmful.

Lung motion sometimes interferes with smooth and rapid progress during chest surgery. Lung motion often reflects tidal exchange. It may be true that even with small tidal exchanges the patient's degree of oxygenation may be satisfactorily maintained. The more difficult problem, however, is satisfactory carbon dioxide elimination. This is dependent upon good tidal volume. Minimal lung movement means unsatisfactory gaseous exchange. As a result, a very large proportion of intrapulmonary surgery is done on patients suffering with marked respiratory acidosis.

Artificial respiration in the operating room for collapse due to overdose of anesthetics or relaxants, too high a spinal anesthesia, trauma, or when carried out for cardiac arrest, can of itself seriously interfere with the patient's chance of recovery, because of the effect of inspiratory positive pressure breathing on cardiac output.

Inspiratory positive pressure breathing of the proper sort may be carried out by manual compression of the bag or by mechanical means. It is our belief that the patient can be better maintained on controlled or assisted respiration by a mechanical device. We have constructed an electronically controlled apparatus which employs oxygen alone, or oxygen in combination with anesthetic gases, and which can produce any desired respiratory pattern. With each of the variables under full control one can produce satisfactory tidal exchange with a low mean pressure (Fig. 5). It can produce controlled respiration alone (Fig. 6), assisted alone (Fig. 7), or a combination of assisted and controlled respiration (Fig. 8). The apparatus can sense an effort by the patient to inspire and it aids him in this regard.

Normal respiration, because of a phasic decrease in intrapleural negative pressure, encourages return of blood to the right heart. Artificial creation of a phase of negative pressure in pressure breathing is becoming increasingly important, for it not only lowers the mean pressure but it aids venous return. This should be of great value in hypotensive states, as in shock. A negative pressure phase may be introduced in the apparatus (Fig. 9).

SUMMARY

Pressure breathing, as fostered by surgeons, is a vital adjunct to present-day surgery and anesthesia. Its tremendous benefits may often be offset by some of its deleterious effects. These effects must be anticipated, recognized,

and treated. To obtain maximum advantage of pressure breathing with a minimum of harmful effects will require the utmost cooperation between the surgeon and the anesthetist.

Please consider this presentation my personal plea to this end.

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CONGENITAL TRACHEOESOPHAGEAL FISTULA WITHOUT ATRESIA OF THE ESOPHAGUS

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ABROAD survey of congenital abnormalities shows that developmental failure is rarely seen in its most severe form, but that lesser degrees of the same fundamental error of development are common. Spina bifida occulta is common; meningomyelocele is rare. Complete exomphalos is rare; umbilical hernia is common. Complete bilateral harelip and cleft palate are far less common than their many minor variants. Many such examples come to mind.

It is surprising, then, that while many hundreds of cases of congenital atresia of the esophagus, with tracheoesophageal fistula, are recorded,[1-3] and very many more are known, but are no longer recorded, there are very few records of minor variants of this congenital abnormality, such as atresia or stenosis of the esophagus without fistula, or fistula without atresia.

There are two possible reasons for this. One is that minor variants occur, but are not diagnosed. The second is that the error of development is peculiar, in that it can give rise only to the full-blown lesion of fistula with atresia, and minor variants cannot occur.

It is the purpose of this paper to suggest that at least one minor variant, fistula without atresia, does occur, but is not diagnosed, and may be far more common than is thought at the present time.

To dispose of the second possibility, it is necessary to review the development of the trachea and esophagus, and to consider the various theories that have been put forward to account for the widely known congenital lesion of fistula with atresia.

EMBRYOLOGY

Vogt's classification of the congenital abnormalities of fistula and atresia is well known, as is the fact that Type III(b) occurs far more commonly than the other variants (Fig. 1). Many theories have been put forward to account for these developmental errors. Pressure by the heart and pericardium, a prominent feature of the developing embryo, has been suggested. The primitive arterial arches, and the great veins, have been incriminated. But none of these "pressure" theories explains the marked preponderance of Type III(b). The most attractive theory is that of Gruenwald,[4] who has described a case of fistula with atresia in a 9 mm. human embryo. He pointed out that normally the trachea is developed as a ventral outgrowth from the foregut, and from that part of the foregut which is to become the esophagus (Fig. 2). It is an embryologic maxim that when a parent tube gives rise to a daughter tube, it does so at the temporary expense of its own lumen, and it is reasonable to assume,

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although this is not known for certain, that during the phase of budding off of the trachea, the esophageal lumen becomes blocked by proliferating cells. When the trachea has separated from the esophagus, the next phase of development is a rapid elongation of the trachea. It elongates not only in proportion to the growth of the embryo as a whole, but in excess of this proportion, so

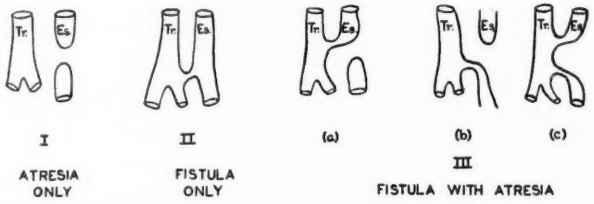


Fig. 1.—Vogt's classification of congenital fistula and atresia. Type III (b) preponderates and is widely reported following surgical correction.

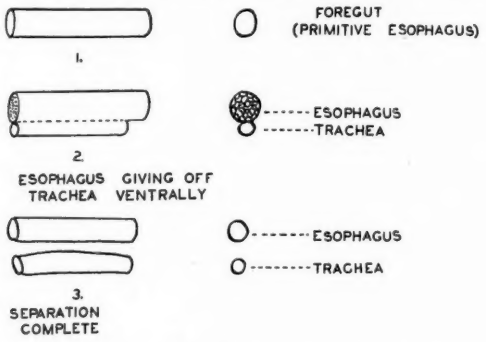


Fig. 2.—Following the separation of the trachea from the esophagus, there is a rapid caudal elongation of the trachea.

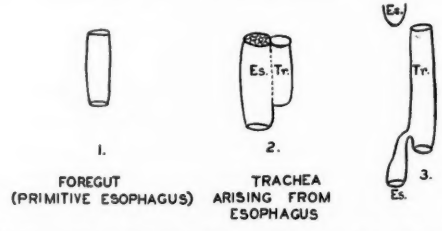


Fig. 3.—The rapid elongation of the trachea takes place before the trachea and esophagus have separated (that is, at Phase 2) and the esophagus is attenuated on the posterior wall of the trachea (3) to produce Type III (b) deformity of Vogt. (After Gruenwald.)

that it elongates relatively to the esophagus, which, of course, is also growing in proportion with the whole embryo. Gruenwald held that the usual anomaly, fistula with atresia, Type III(b), is due to the failure of coordination of the two phases of development, so that the rapid elongation of the trachea takes place before the trachea becomes separated from the esophagus (Fig. 3). In this event, the esophagus, its lumen probably blocked by cells, will be drawn out along the posterior wall of the trachea as an attenuated strip, or as a thin cord,

or fragmented. The site of the fistula between the lower end of the esophagus and the trachea will depend upon the degree of separation between the two at the time that caudal elongation of the trachea takes place. If this rapid growth takes place at an early stage, then the fistula will be near the carina, but if separation is almost complete, then the fistula will be high in the trachea.

Gruenwald considered that the upper blind pouch is produced by the growth of a part of the primordial esophagus attached to the pharynx. His theory is reinforced by the discovery of squamous epithelium and striated muscle in the posterior wall of the trachea in cases of fistula and atresia.[5-7] Rosenthal[8] has observed a case in which the esophagus was visible as a longitudinal thickening on the posterior wall of the trachea. I have observed, in cases of patients with fistula with atresia on whom I have operated, that there is a clear angle between the trachea and esophagus below the fistula, but that above it, there is often a band or cord running up the posterior wall of the trachea for a variable distance, making the fistula difficult to define at this point. Many other surgeons performing these operations must also have observed this, but its significance becomes apparent only with the background of Gruenwald's theory.

Gruenwald pointed out that esophageal remnants may not be found in the posterior wall of the trachea. This does not affect the validity of his theory, since it is well known that intercalated structures, such as grafts, may be incorporated in the final structure of an organ, in so far as their fundamental structure allows transformation under the influence of the anlage of the developing parent organ.

Gruenwald's theory has the beauty of simplicity. It explains why the tracheoesophageal fistula almost always involves the lower end of the esophagus, and why atresia is almost always accompanied by fistula. But it does not explain why fistula should not occur without atresia, and indeed one would expect that since the trachea is developed from the esophagus, congenital fistula between them would be common. If Gruenwald's theory is correct, there is nothing in it showing that fistula without other abnormality cannot occur.

If it be granted that, on embryologic grounds, fistula without atresia can occur at least as readily as fistula with atresia, then one asks why the condition is not met more frequently in clinical practice. There are three possible answers to this. First, fistula with atresia was considered a rare condition until a surgical cure was devised, and simple diagnostic measures were widely published. Second, the simple diagnostic measures which prove the presence of fistula with atresia will not necessarily show the presence of a fistula only. Third, the clinical features of fistula without atresia lack the dramatic quality of fistula with atresia; they do not necessarily suggest the diagnosis in themselves; and the onset of symptoms may be delayed until adult life. These latter points are brought out by a survey of the literature of cases of fistula only, and by the two new cases now recorded.

REVIEW OF LITERATURE

The literature of congenital abnormalities of the esophagus, including tracheoesophageal fistula, was extensively reviewed by Cautley[9] in 1917. A

further review was made by Plass[10] in 1919, prompted by a case of fistula with atresia, and again more recently by Haight[11] in 1948, prompted by the discovery of two cases of fistula without atresia. Many of the cases here reviewed have been noted by one or the other, or all three, of these authors.

The first recorded case of fistula only is that of Richter,[12] in 1792. It was a peculiar abnormality in which the trachea and esophagus formed a single sac. This can be classified as a fistula in its most gross form, but otherwise has little similarity to the other cases. One can see, however, how such an abnormality might arise on the basis of Gruenwald's[4] theory.

In 1849, Vrolik[13] recorded a case, classified by Cautley[9] as one of fistula without atresia, but by Plass[10] as one of fistula with atresia. Unfortunately, the original paper is not available in this country.

Cautley[9] mentioned Von der Water[14] as recording the next case in 1857, but once again, the original reference is unobtainable.

In 1873, Lamb[15] recorded a case in an American journal. Haight[11] has verified this case, which, he stated, was of a patient who died at the age of 7 weeks. In the same year, 1873, Pinard[16] and Tarnier[17] independently recorded the same case, which Haight[11] has again verified, and which he stated was a high subglottic fistula, similar apparently to that successfully closed by Imperatori.[29] Pinard and Tarnier's patient died at 36 hours.

Eppinger[18] recorded a case of fistula, just above the carina, in a "young man" who died. Haight[11] has verified this case also, and it is the first case recorded in the literature, with reasonable certainty, of a congenital fistula being found in an adult. This was in 1880.

Heiderich,[19] at a demonstration of pathologic anatomy in 1916, showed a specimen of human thoracic viscera which showed a cord joining the esophagus to the right bronchus. The cord, which could be easily lifted out, and had no diseased lymph glands around it, started with a broad base in the esophagus, some of whose longitudinal fibers turned into it. The other end was attached to the posterior wall of the right bronchus some 2 cm. from the bifurcation. The esophagus, when opened, revealed a funnel-shaped pouch corresponding to the place where the cord started. It slanted downward in a short canal, which easily allowed a fine probe to pass, and followed the axis of the cord, finally opening into the right bronchus with a small round opening. Heiderich believed that this fistula was congenital. It would appear that the congenital origin was accepted in the discussion. In 1917, Cautley[9] made his extensive review, already mentioned. His paper consists of a classification of the recorded abnormalities of the esophagus, and included six cases of fistula without atresia, those of Richter,[12] Von der Water,[14] Lamb,[15] Pinard[16] and Tarnier,[17] Vrolik,[13] and Eppinger.[18]

In 1919, Plass[10] followed with a further review. His paper is of interest in that it quotes verbatim the first account, by Thomas Gibson in 1696, of a case of fistula with atresia. Plass quoted Lamb[15] as his only case of fistula without atresia. He did not include Eppinger[18] or Von der Water[14] in his review at all. He quoted Pinard[16] and Tarnier[17] as a case of fistula with atresia, but Haight,[11] as stated previously, has verified this case as one of fistula only.

In 1921, Widmann[20] gave a very full account of a post-mortem on a 34-year-old "Landstrumm" recruit, who died following an acute exacerbation of chest disease of fourteen years' duration. The terminal illness was in June, 1918, characterized by fever, headache, and bloody sputum. Death occurred in July, following a severe hemoptysis. Autopsy showed an old "obsolete" lesion at the left apex, with caseous softening of the lymph glands at the hilus. There was old bronchiectasis of the left lower lobe, with "callous thickening of the stroma." The left lung was densely adherent. There were sanguinopurulent bronchitis and bronchopneumonia of the left lung. On the right side, there were light adhesions. Finally, there was a tracheoesophageal fistula, situated on the posterior wall of the trachea, where it began as a funnel as thick as a pencil, diminishing to a few millimeters wide in the middle of the posterior wall at the level of the tenth cartilaginous ring. It passed steeply backward and downward through the anterior wall of the esophagus, entering the lumen 7.7 cm. below the interarytenoid indentation. The lining of the fistula was free from scarring, and admitted a 2 mm. probe throughout. There was no surrounding scarring or lymphadenitis. In the discussion which followed the presentation of this case, which was accepted as congenital, it was agreed that cases of fistula without constriction of the esophagus were rare. Stubler described one case and referred to three others, the patient in one of which survived seven weeks (possibly Lamb's case). The long survival in Widmann's case was attributed to the direction of the fistula, downward from the trachea to the esophagus.

Although so few cases had been recorded up to that time, reference is made to the condition in two textbooks published in 1928 and 1929. Anders[21] stated, "In the adult, tracheoesophageal fistulas, which can sometimes be very fine, can give passage to particles of food into the trachea, and they can cause aspiration pneumonia or gangrene of lung." Jackson and Coates[22] stated, "Congenital fistulae are usually fatal; the patients who survive are those with only a small opening covered by a fold or a thin film of membrane. In one such patient who came to the Bronchoscopic Clinic, the film had evidently given way two years before; fatal pulmonary complications had been prevented by feeding with a stomach tube."

In the same year, 1929, Negus[23] showed to the Royal Society of Medicine a specimen of the esophagus of a man of 45 years, who had died from carcinoma of the lung. There was a congenital opening, passing downward from the trachea to the esophagus. At the esophageal end was a valvelike hood of mucous membrane. There had been no symptoms during life. Negus has seen no other case of this condition, and unfortunately the specimen was destroyed during the war.[24]

Clerf[25] reported, in 1933, a case of a woman of 23 years, in whom a fistula between the esophagus and the left lower lobe bronchus was demonstrated by esophagoscopy, barium swallow, and radiography following the passage of radiopaque bougies. The state of the lung is not recorded in any detail. The fistula was treated by cauterization with silver nitrate, and appeared to obli-

erate. The fistula was 29 cm. from the upper teeth at esophagoscopy. No opinion was given on the cause of this fistula, but the history was of seven years' coughing on swallowing. The patient's age at onset was thus 16 years, and since no other disease is mentioned, it is reasonable to assume a congenital origin. Late progress is not reported.

In the same year, McKinney[26] reported another case in the same journal. This was a woman of 58 years, with a history of recurrent attacks of vomiting for twenty-five years, and with recent attacks of coughing and spitting up food after meals. The fistula was shown by barium swallow, and confirmed by bronchoscopy when beads of pus could be seen passing through the fistula, which was itself too small to be seen. Esophagoscopy failed to show the fistula, too small to be found among the folds of mucosa. A further barium swallow confirmed the previous findings. McKinney reviewed the literature at the time, and considered that it was a congenital fistula, occluded until middle life by a membrane. He reported a personal communication from Chevalier Jackson, in which Jackson stated that he had found a number of such fistulas in older children and adults.

Two, and possibly three, cases were reported in 1937. Moersch and Schmidt[27] reported two cases. The first was of a woman of 31 years, who gave a history of coughing up particles of food for eight years. A fistula passed from the lower esophagus to the right lower lobe bronchus. There was suppurative pneumonia of the right lower lobe. Death followed an operation, the details of which are not given. Autopsy showed no cause for the fistula, which was large. It was seen at esophagoscopy and bronchoscopy. Moersch and Schmidt's second case was of a man of 52 years, whose history was of strangulation and dyspnea on drinking, since childhood. Solids caused little distress. There had been four attacks of pneumonia in the left lower lobe. The fistula passed to the left lower lobe bronchus; it was seen at esophagoscopy, when the orifice was observed to open and close, and could easily have been missed if closed. The authors regarded their second case as congenital, the etiology of the first being in doubt.

In 1937 also, Tobias and Carlone[28] reported a case of "congenital esophagotracheal fistula in a man of 35, complicated by a gangrenous abscess of lung." This article is not available in this country.

In 1939, Imperatori[29] described a case of fistula in a boy of 6 years. The fistula was high, at the level of the third and fourth tracheal rings, and was repaired by a transcervical, transtracheal route. This case appears to be similar to that of Pinard[16] and Tarnier.[17]

In 1941, Gengenbach and Dobos[30] reported ten cases of fistula, the patients of seven of which had atresia. Of the remaining three, one was of fistula without any other abnormality of the esophagus, and was proved at autopsy in a child of 3 months. The fistula was 4 mm. in diameter, and passed downward from the trachea to the esophagus, entering the trachea 3 mm. above the carina. The second case was of a child of 10 months, diagnosed by bronchoscopy. There

was some obstruction or stenosis of the esophagus. This child was alive at the time of the report. The third case was suspected, the diagnosis not being proved at the time of the report.

In 1945, Leven and Lannin[31] discussed cases of fistula with atresia, and in the course of the article, mentioned two cases of fistula without atresia. These cases were found in the records of the Department of Pathology of the University of Minnesota, between 1916 and 1938. No details are given.

Haight,[11] in 1948, recorded a case in a child of 4 years. There had been choking on feeding since birth and, soon after birth, fistula with atresia was suspected. The esophagus was shown to be patent by barium swallow, on the fourth day of life. The barium swallow did not outline the fistula. Attacks of pneumonitis persisted, and the diagnosis was made at the age of 3 years, by Lipiodol swallow in the prone position. Bronchoscopy showed the fistula 1.5 to 2 cm. above the carina, in the right posterolateral wall of the trachea. The fistula was 4 mm. in diameter, at its narrowest. Haight successfully closed the fistula by an extrapleural paravertebral approach. Haight had previously seen a similar case, which he had reported in a paper on fistula with atresia,[32] and which is also described in Patten's textbook of embryology.[33] The radiologic features of fistula with atresia were dealt with by Haight and others in a further paper.[34] He pointed out that the usual procedure for establishing the diagnosis of fistula with atresia, namely, to instill a little Lipiodol into the upper esophagus through a fine catheter, will not outline a fistula if the esophagus is patent. The oil will run into the stomach, and will pass through the fistula into the trachea only if it is instilled into the esophagus in the prone position. In his paper[32] Haight stated that he is aware of two other cases of fistula only, which do not appear to be reported in the literature.

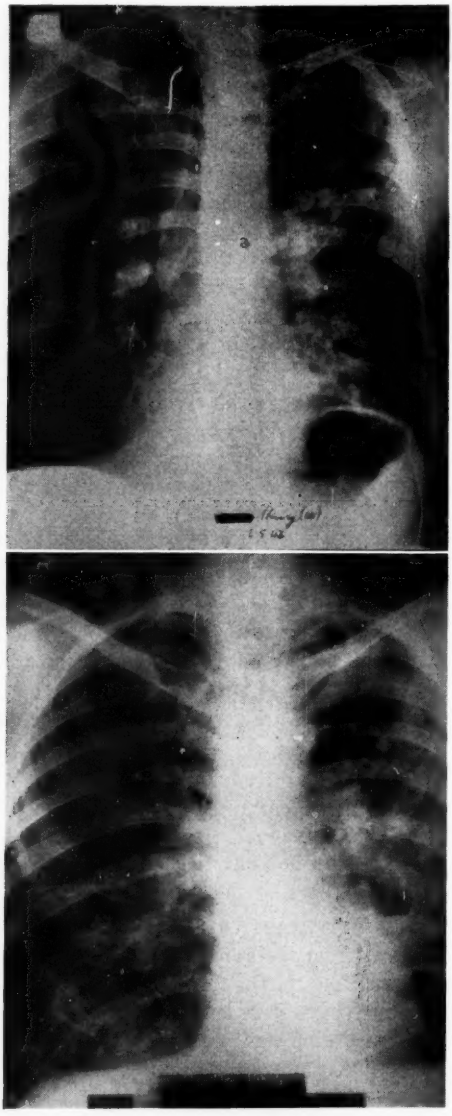
To retain the chronological order of this review of case reports, my own first case must be mentioned at this point. This case was diagnosed and treatment was made in 1948, and it was reported to the Society of Thoracic Surgeons of Great Britain in 1949. Details are given in the case reports in the following section.

Morton, Osborne, and Klassen recorded a case in 1950.[35] The patient was a man of 26 years, in whom a congenital fistula passed from the esophagus downward to the right lower lobe bronchus. The symptoms of coughing of ingested food particles did not arise until adult life. He was successfully treated by right lower lobectomy and closure of the fistula.

Berman, Test, and McArt[36] reported a case in 1952 of a woman of 40 years in whom the fistula passed from the esophagus to the left lower lobe bronchus. Symptoms had been present from birth. She was treated by left pneumonectomy and closure of the fistula. There were associated vascular abnormalities in this case. A patent ductus arteriosus was present in the usual position, and there was a second ductus, arising from the aorta and communicating with the basal branch of the pulmonary artery. This suggests that the developmental error may have been of a different kind from that postulated for these cases, and possibly of the kind which gives rise to "sequestration of lung," a condition

described by Pryce, Sellors, and Blair[37] and now familiar to most thoracic surgeons. Berman, Test, and McArt mentioned four cases of fistula without atresia, of congenital origin, in their article,[36] of which two are not included in this review. These are recorded by Walters[38] and Berglund.[39]

A.



B.

Fig. 4.—Case 1: Radiographs showing pneumonitis in the left lung in 1942 (*A*) and 1948 (*B*). In the later film, there is a well-marked cavity in the left lower lobe, and a pneumonitis in the right mid-zone.

A further case has recently been recorded by Krausey,[41] in 1953, in an African woman of 38 years. The fistula was very large, 4.4 by 1.9 cm., and death resulted from asphyxia, following impaction in the fistula of a bolus of meat.

There are thus some thirty cases of congenital esophagotracheal or esophagobronchial fistula recorded in the literature, about equally divided between adults and children. Three of these have been treated by transthoracic operation. To this list I am able to add two further cases.

Fig. 5.—Case 1: A barium swallow, showing barium entering the left main bronchus.

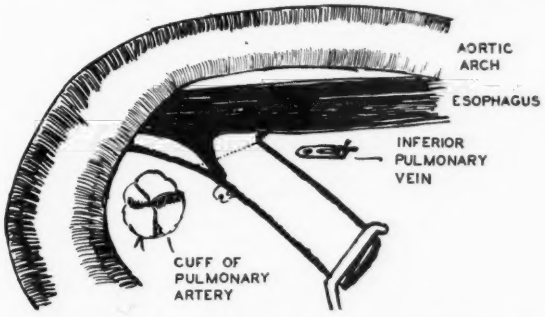


Fig. 6.—Case 1: Drawing of appearances after pneumonectomy. The bronchial stump has been left long, and is being used to draw the fistula from beneath the aortic arch. The subsequent line of section of the bronchus is shown dotted.

CASE REPORTS

CASE 1.—A 54-year-old man had a history of cough with purulent sputum for eleven years, and of coughing of ingested food particles for six months. He had been investigated in many hospitals, and had been in a sanatorium for three months. No tubercle bacilli had ever been found in the sputum. Two years previously, a bronchogram had suggested the presence of a fistula, but esophagoscopy and bronchoscopy had failed to reveal it. A survey of the radiographs taken over many years showed a chronic infective process in the left lung,

mainly in the lower lobe, in which there was a cavity. (Fig. 4.) There had been many exacerbations and remissions of the infective process, but the right side had suffered only a transient pneumonitis in 1948. The development of the pathognomic symptoms of coughing

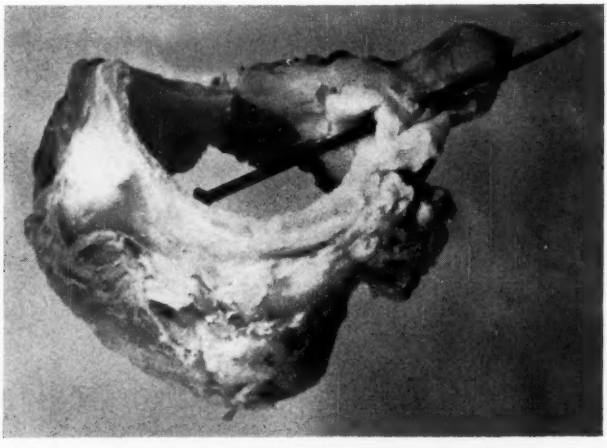


Fig. 7.—Case 1. The segment of the left main bronchus, showing the fistula.

Fig. 8.—Case 1. Sections of the resected left lung.

of ingested food particles, symptoms which had not been present two years previously, when a fistula had been suspected on a bronchogram at a previous investigation in another unit, now left the diagnosis in little doubt. A barium swallow showed the fistula, passing from the esophagus to the left main bronchus. (Fig. 5.) The mouth of the fistula could not be

seen at esophagoscopy, and was only found with difficulty at bronchoscopy. This was because of the extremely small bronchial mouth, situated in the depression between two cartilaginous rings. A probe could just be passed into it.

On June 17, 1948, left pneumonectomy and closure of the fistula were carried out. The chest was opened through the bed of the resected sixth left rib, the patient being in the prone position described by Parry Brown.[40] The lung was densely adherent to the chest wall and diaphragm, but the hilum and mediastinum were not involved in this fibrosis. Dissection pneumonectomy was performed, leaving the stump of the bronchus long, to act as a tractor to draw the carina and fistula from beneath the aortic arch. This was done with surprising ease, and the fistula displayed with a minimum of blunt dissection. (Fig. 6.) The fistula entered the posterolateral wall of the left main bronchus 1.5 cm. below the carina at the junction of the cartilaginous and posterior membranous portions of the wall of the bronchus. It was 2.3 mm. outside diameter at the point of entry into the bronchus, and about 1 cm. in length. Funnel shaped, it widened rapidly so that it was 0.75 cm. diameter at its entry into the esophagus, and about 1 cm. in length. The bronchus was clamped above the fistula, and divided. The fistula was divided at its entry into the esophagus, and the

Fig. 9.—Case 2. A barium swallow, showing the fistula passing to the right dorsal lobe bronchus.

segment of bronchus, with the fistula, was removed. (Fig. 7.) The defect in the esophagus was closed with a few interrupted linen thread sutures, and the bronchus closed with interrupted linen thread, and covered with a pedicled intercostal muscle graft. The chest was closed without drainage. The patient made a good recovery, and returned to work, but in February, 1949, eight months after operation, he made a violent effort to lift a heavy weight, and felt a sharp pain in the chest. Within twenty-four hours, he was admitted to the hospital, with fever and a profuse productive cough. He had in fact developed a virulent empyema, due to rupture of the bronchus stump. The empyema was drained, and he left the hospital, to return within a few days after a severe hemorrhage from the empyema drainage tube. Hemorrhages of increasing severity recurred during the following few days,

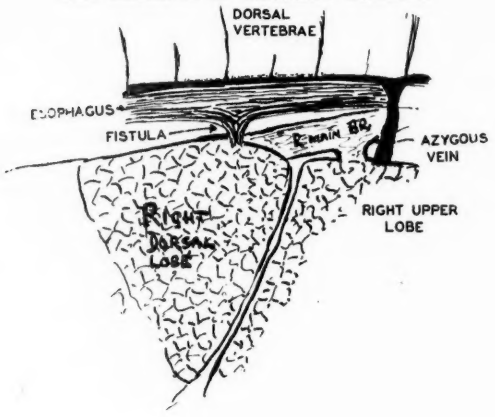


Fig. 10.—Case 2. Diagram of appearances at operation.

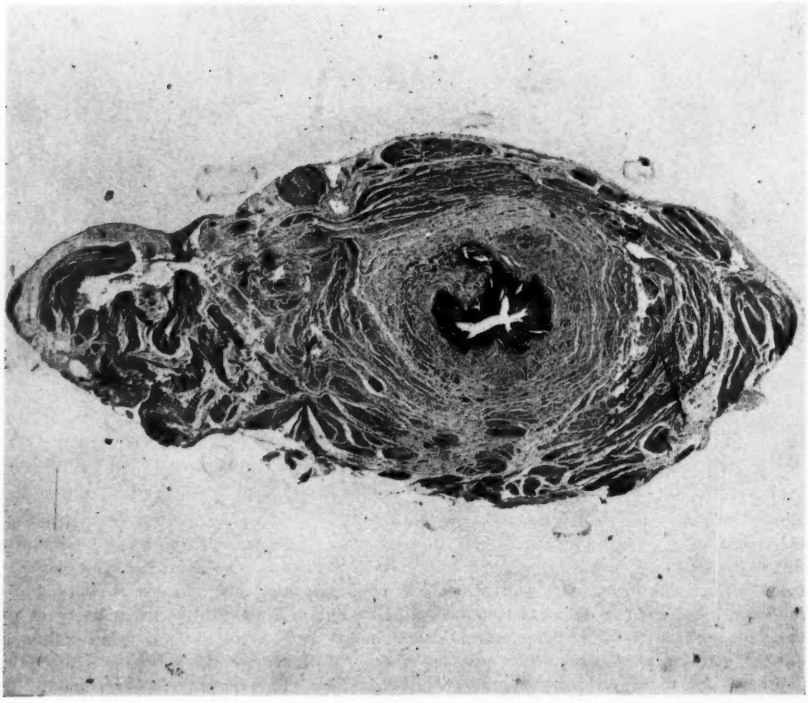


Fig. 11.—Case 2. A photomicrograph of a cross section of the fistula, showing a muscular wall, and a squamous epithelial lining.

and the chest was reopened in order to control the hemorrhage. As soon as this was done, a torrential hemorrhage took place, and he died within a few seconds. Post-mortem showed that the hemorrhage was due to rupture of an infective aneurysm of the descending arch of the aorta. This had converted the pneumonectomy space into a false aneurysmal sac. The post-mortem showed no evidence of disease which might have caused the broncho-esophageal fistula. Examination of the resected left lung had shown only chronic nonspecific inflammatory changes, with no evidence of tuberculosis, actinomycosis, or neoplasm. (Fig. 8.)

CASE 2.—A 54-year-old woman gave a history of enlargement of the thyroid gland for two to three years; one year before coming to the hospital, she had noticed a bad taste in the mouth, and for five months she had suffered from regurgitation of fluids from the esophagus, which had become worse in the previous two months. There was no clear-cut history of coughing on swallowing. The symptoms were attributed to the enlarged thyroid, and thyroidectomy was carried out uneventfully, but without relief of symptoms. A barium swallow was carried out, and this revealed a fistula passing from the esophagus to the right dorsal lobe bronchus. (Fig. 9.) She was then referred to the Thoracic Unit. Esophagoscopy showed a suggestion of the fistula 27 cm. from the upper incisor teeth, a dimple in the mucosa. At bronchoscopy, there was likewise a suggestion of a fistula in the floor of a groove in the posterior wall of the right main bronchus, just above the dorsal lobe orifice. Except for the unequivocal demonstration of the fistula by barium swallow, its presence would not have been suspected at these endoscopies. On Jan. 23, 1953, a right thoracotomy was performed with the patient in the prone position. The dorsal lobe had a well-marked fissure separating it from the basal segments, and there were filmy adhesions over the dorsal lobe. The fistula could be seen through the mediastinal pleura. There was no inflammatory reaction around it, and it was picked up and dissected out without difficulty. It arose from the lateral aspect of the esophagus, and ran at right angles to it, to enter the medial side of the dorsal lobe bronchus, just distal to its origin from the main bronchus. It was funnel shaped, about 2 cm. in length, 1 cm. broad at the esophageal attachment, and 0.5 cm. broad at its bronchial attachment. (Fig. 10.) It was ligated at its bronchial attachment, and divided. The esophageal attachment was divided, and the esophageal defect closed with a few interrupted linen thread sutures. The chest was closed without drainage. The patient made an uneventful recovery, with complete relief of symptoms. The fistula, after excision, easily admitted a probe; it was lined with squamous epithelium. (Fig. 11.)

DISCUSSION

Of the thirty-odd cases now reported, no clinical details whatever are available for six of them.[12-14, 19, 31] In another nine, only the age at diagnosis or death is known; of these nine, four patients died within a few days of birth[11, 15-17]; another died at age 3 months[30]; two cases were diagnosed during the first year, and the patients were living when the cases were reported[30]; and the other two were adults, one a "young man,"[18] the other aged 35 years.[28] Fifteen cases remain, concerning which clinical details are known. One patient died aged 2 or 3 days, of pneumonia[11]; another was aged 23 months, and had lung symptoms[38]; a third had recurrent attacks of pneumonitis, and was successfully operated upon at the age of 4 years, by Haight.[11] A fourth had an unusual type of fistula, high in the subglottic region, and a difficult history of regurgitation, complicated by a history of pyloric stenosis. This fistula was successfully closed by Imperatori[29] in a boy of 6 years.

Haight has given a very full account of the clinical aspects of the condition in childhood and infancy, and the present discussion will be confined to the remaining eleven cases, in which the abnormality was first diagnosed in adult life, and in which there is available an adequate record of the case history. The

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striking fact immediately emerges from these eleven case histories that in only two of them did symptoms date from birth or early childhood. One patient had no symptoms referable to the fistula; in the other eight, symptoms first occurred in adult life. Furthermore, symptoms of coughing of ingested food were present from the onset in only five of the eleven, and were not found at all in two. These facts are shown diagrammatically in Fig. 12. This is important, because it leads to difficulty in diagnosis in the early stages, when the patient presents with recurrent or chronic attacks of pneumonitis; unless a fistula is suspected, it is very unlikely to be found accidentally, and even if suspected, proof of its presence is not easy to find, since the very small orifices of the fistula in bronchus and esophagus make it very difficult to see them, even when their approximate location is known, and when diligent search is being made; also, esophagoscopy

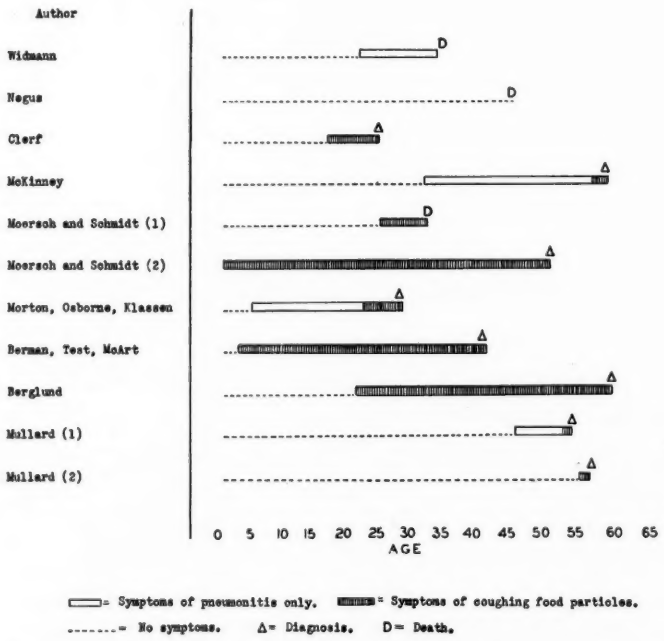

Fig. 12.—Diagram showing age at onset and duration of principal symptoms in adult patients.

does not often form part of the investigation of cases of recurrent pneumonitis. The key investigation is an opaque swallow, using either a thin barium mixture or Lipiodol. Swallows should also be carried out in the prone position, so that gravity can assist the opaque material to flow forward into the trachea or bronchus. Although it is improbable that these fistulas are at all common, it may well be that they are a good deal more common than the published reports suggest, and it may be advisable to include an opaque swallow in the investigation of cases of pneumonitis which are recurrent or refractory to treatment.

Many of the fistulas described, including the one in my Case 1, have passed from the esophagus to the trachea above the carina. It would be possible to carry out lobectomy, and even pneumonectomy, without discovering the presence of the fistula, which in these cases lies entirely within the upper mediastinum, in an area undisturbed during pulmonary resection. If resection of a destroyed lobe or lung were carried out in the presence of such a fistula, which remained undetected, then one would anticipate progressive infection in the residual lobe, or in the contralateral lung. It may be that this is the explanation of some of those cases of progressive pulmonary suppuration following resection, which occur occasionally in the practice of most thoracic surgical units.

The reasons for the late onset of symptoms in general, and of the pathognomic symptoms in particular, are worth consideration. Jackson and Coates[22] considered that the fistula is occluded by a fold or membrane which breaks down in later life. While this is possible, there has been no evidence of such a membrane in my cases, in both of which the fistula has been lined with squamous epithelium, continuous from esophagus to bronchus. The esophageal mucosa is normally mobile within the muscular coat, and moves downward with the bolus on deglutition. But the mucosa is firmly attached to the fistula, and this will cause a fold of mucosa to develop and override the orifice of the fistula as the bolus passes, giving some protection against entry of food particles into the fistula. This mechanism will also account for the difficulty of seeing the orifice of the fistula at esophagoscopy. But there is no reason why this mechanism should fail in course of time.

Another possibility is that the direction of the fistula, upward or downward from the esophagus, accounts for the variation in the time of onset of symptoms from one case to another. There is insufficient evidence in the case reports to support this theory.

It may be that the explanation of the late onset of symptoms in many cases is that the normal mobility of the esophagus in relation to the other mediastinal contents, and especially the bronchi, is interfered with by the progressive adherence of the affected lung to the chest wall and mediastinal pleura. Adhesions form, and become increasingly dense, with each of the recurrent attacks of pneumonitis. Eventually the affected lobe or lung will become firmly adherent to the chest wall and mediastinal pleura. The esophagus normally descends on deglutition, and the esophageal mucosa descends in relation to the muscular coats. When the lung becomes fixed, the free descent of the esophagus will be restricted, at least at the point of attachment of the fistula. The tendency may well be that the fistula is pulled open by this means, while at an earlier stage, when there was free movement of the lung, bronchus and fistula, as well as esophagus, descended together, the mouth of the fistula being occluded by a fold of esophageal mucosa during the passage of the bolus, as described previously.

SUMMARY

The embryology of congenital tracheoesophageal fistula, with and without atresia of the esophagus, is reviewed.

The published reports of congenital fistula without atresia are summarized. Two new cases of this condition are reported, both diagnosed during life and treated by direct transthoracic approach.

The clinical history and symptoms of cases of fistula without atresia, with special reference to the condition in adult life, are discussed.

Attention is drawn to the late onset of symptoms in most cases, and especially to the late onset of pathognomic symptoms, and also to the difficulties of diagnosis.

A theory is advanced to account for the late onset of symptoms.

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POSTINFLAMMATORY "TUMORS" OF THE LUNG

Report of Four Cases Simulating Xanthoma, Fibroma, or Plasma Cell Tumor

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INTRODUCTION

IT IS the purpose of this paper to present four cases with pulmonary lesions characterized by mild or no symptoms, roentgenographic pattern of a single sharply circumscribed mass suggesting tuberculoma or neoplasm, histologic features of fibroma, neurofibroma, xanthoma, or plasma cell tumor, and historical evidence of an antecedent respiratory infection.

Brown and Johnson[1] recently reported three such tumorlike lesions of the pleura, which they regarded as consequences of inflammation. They stated that similar masses of inflammatory origin had been found within the lung.

REPORT OF CASES

Case 1†.—A 24-year-old white man was being examined prior to separation from the Navy, when a small intrathoracic mass was seen in roentgenograms of the chest (Fig. 1). Roentgenograms taken four months earlier had been negative, but for the previous two months, he had had a slight cough productive of small quantities of yellow sputum. Extensive bacteriologic and cytologic studies did not reveal the cause of the lesion, which was tentatively diagnosed "pulmonary tuberculoma." Right upper lobectomy was performed.

A well-circumscribed, soft, spherical nodule, 3 cm. in diameter, was found 2 cm. beneath the pleura. The central cystic space was surrounded by a friable, granular, yellowish brown wall. Microscopic sections of the mass showed bundles of interlacing spindle-shaped cells with relatively abundant acidophilic cytoplasm and large nuclei (Fig. 2). The nuclei were rather variable in size and shape, and some were moderately bizarre, yet no mitotic figures were found. In other areas, numerous macrophages were roughly polyhedral, with round or oval nuclei (Fig. 3). Some contained brownish pigment that stained positively for iron. Clusters of foam cells containing sudanophilic fatty material were a striking feature, especially near foci of hemorrhage (Fig. 4). Scattered throughout were varying numbers of chronic inflammatory cells, mostly plasma cells, with fewer lymphocytes and neutrophilic polymorphonuclear leukocytes (Fig. 5). The adjacent pulmonary parenchyma was the site of recent hemorrhage.

Case 2‡.—A 5½-year-old white girl became ill, in February, 1937, with moderately productive cough, fever, nausea and vomiting, sore throat, and cervical lymphadenopathy. The acute symptoms subsided in five days, leaving some weakness, slight clubbing of the fingers, and daily elevation of temperature. Roentgenograms one month later revealed a large, annular mass in the right lung (Fig. 6). Three months following the onset of illness, a right upper lobectomy was performed.

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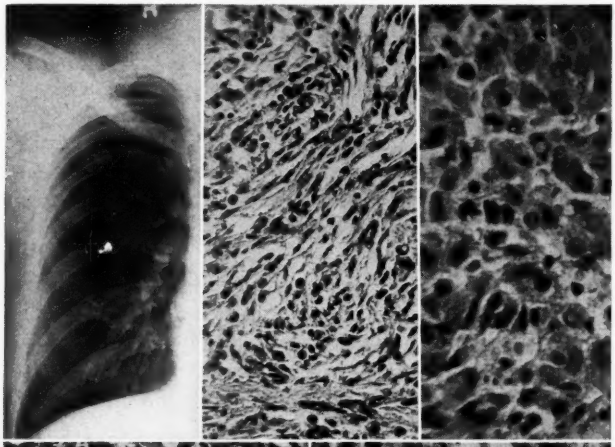
‡AFIP Acc. No. 170825, contributed by the late Dr. H. Brunn,[2] San Francisco, California.

The well-circumscribed, soft, spherical mass was 5 cm. in diameter. The cut surface was pink tan and somewhat gelatinous, with areas of hemorrhage and strands of fibrous connective tissue. Microscopic sections showed the "tumor" to be partly composed of spindle cells, associated with a loose diffuse network of adult fibrous connective tissue.

Fig. 1. Fig. 2. Fig. 3.



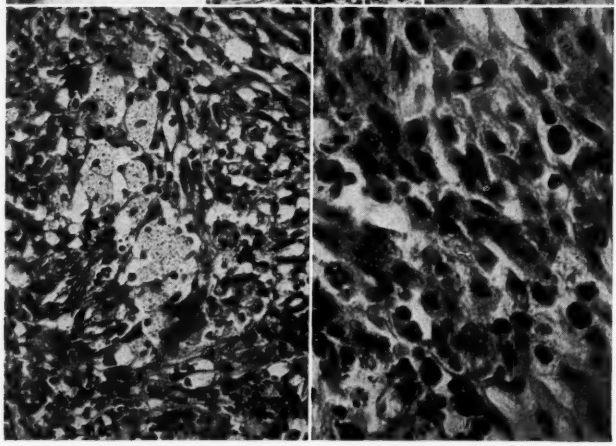


Fig. 4. Fig. 5.

Fig. 1.—Case 1: Roentgenogram of the chest showing a sharply circumscribed mass in the right upper lobe.

Fig. 2.—Case 1: The "tumor" is composed of interlacing bundles of spindle cells with scattered plasma cells and lymphocytes. (×250.)

Fig. 3.—Case 1: Macrophages and scattered small inflammatory cells. (×475.)

Fig. 4.—Case 1: Aggregates of foam cells, as seen here, are not uncommon. (×425.)

Fig. 5.—Case 1: Plump spindle cells and plasma cells are hall marks of this "inflammatory tumor." (×635.)

The bulk of the mass was composed of plasma cells, mononuclear cells, and lymphocytes (Figs. 7 and 8). No mitotic figures were seen. The patient was living and well seven years after operation, and there had been no x-ray evidence of recurrence.

CASE 3*.—A 35-year-old white woman with a history of repeated attacks of bronchitis complained of vague pain in the chest for seventeen months. On roentgenographic examination in 1943, she was found to have a radiopaque shadow in the left lung, which was tentatively diagnosed as tuberculoma. Roentgenograms taken regularly during the following years showed the mass unchanged except for more obvious circumscription. It was removed by lingulectomy at the end of six years.

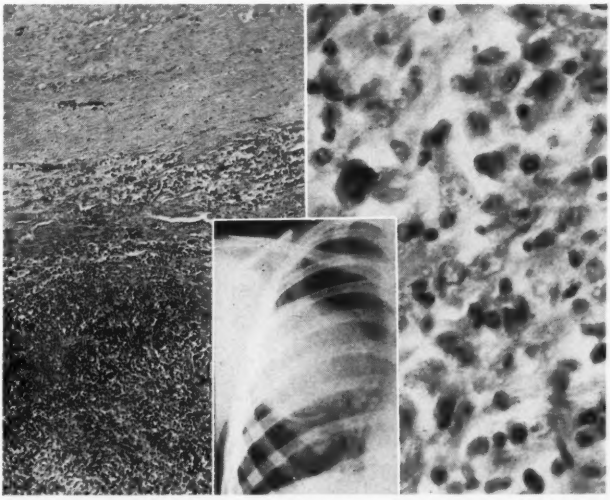


Fig. 7. Fig. 6. Fig. 8.

Fig. 6.—Case 2: Roentgenogram of the chest showing a large mass involving the right lung.

Fig. 7.—Case 2: The fibrous capsule surrounding a dense accumulation of cells, many of which are plasmacytes. (×75.)

Fig. 8.—Case 2: Spindle and polyhedral cells are interspersed with plasma cells, some of which are binucleated. (×555.)

The encapsulated nodule was 2.5 cm. in diameter (Fig. 9). Its cut surface was smooth and golden yellow, with several hemorrhagic and firm, translucent foci. Microscopic examination revealed a thin fibrous capsule enclosing a mass composed of bundles of spindle cells that appeared to be laying down collagenous connective tissue (Fig. 10). The bulk of the tumor consisted of large macrophages with foamy or granular cytoplasm (Fig. 11). Although the nuclei of the spindle cells were somewhat variable in appearance, there were no mitotic figures. Scattered plasma cells were observed (Fig. 12). Focal hemorrhages were present, and aggregates of lymphocytes were seen at the periphery.

CASE 4†.—A 41-year-old white man had "lobar pneumonia" in 1928, when he was 19 years old, followed by a persistent cough productive of brownish sputum. The respiratory symptoms continued for twenty years. In 1948, he was thought to have an adenoma of the bronchus of the lower lobe of the right lung. The obstructing mass was removed bronchoscopically with temporary relief of symptoms. Bronchoscopic examination in 1950, twenty-two years after the initial illness, disclosed a mass obstructing the middle and lower main bronchi on the right. Lobectomy was performed shortly thereafter.

The encapsulated, spherical mass measured 7 cm. in diameter. The cut surface bulged and was light yellowish gray with small yellow flecks. Microscopic examination showed

*AFIP Acc. No. 280217, contributed by Dr. Thomas Peery, Washington, D. C.; reported by Ford, Thompson, and Blades.[3]

†AFIP Acc. No. 485798, contributed by Dr. C. Allen Payne, Grand Rapids, Michigan.

interlacing bundles of spindle cells and hyalinized fibrous connective tissue. Although these cells varied in size and shape, and possessed atypical nuclei, no mitotic figures were seen (Fig. 13). Interspersed among these elements were scattered plasma cells and lymphocytes in varying numbers (Fig. 14). Nests of foam cells were present. The adjacent lung tissue exhibited some fibrosis, pigmented macrophages, and a few lymphocytes. In the year that elapsed after operation, there was no evidence of recurrence.

Fig. 9. Fig. 10.

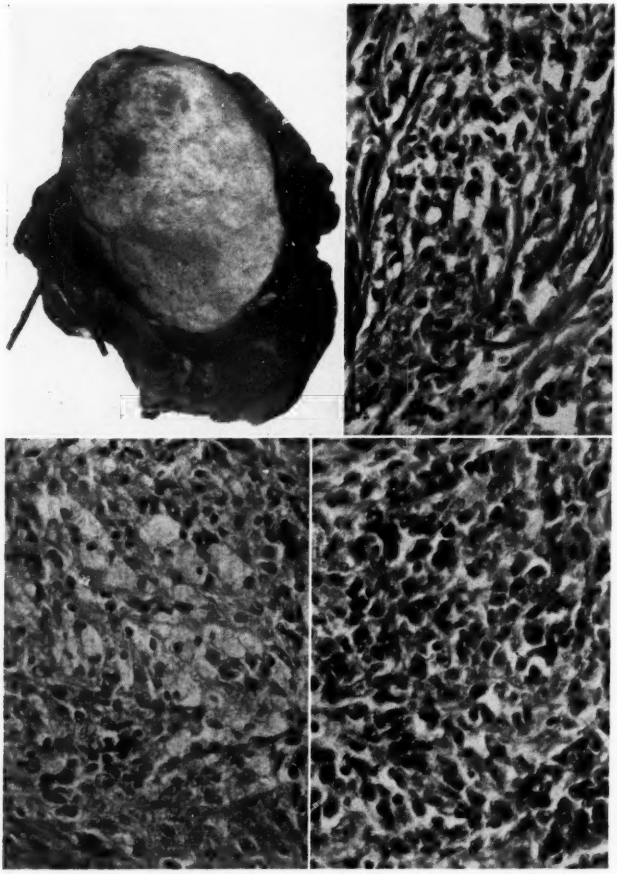


Fig. 11. Fig. 12.

Fig. 9.—Case 3: This well-encapsulated mass was golden yellow. A hemorrhagic focus is seen on the left side. (×3.)

Fig. 10.—Case 3: Hyalinized fibrous connective tissue fibers and scattered inflammatory cells, most of which are plasma cells. (×435.)

Fig. 11.—Case 3: Foam cells containing fat contributed to the yellow color of this "tumor." Spindle, polyhedral, and scattered inflammatory cells are also seen. (×355.)

Fig. 12.—Case 3: This area of the "tumor" contains numerous plasma cells. (×435.)

TABLE I. SUMMARY OF CLINICAL AND PATHOLOGIC FEATURES OF SEVEN INFLAMMATORY TUMORS OF LUNG

AUTHOR	SEX AGE	LOCATION OF MASS	CHIEF SYMPTOM	PAST HISTORY	CLINICAL DIAGNOSIS	GROSS APPEARANCE	MICROSCOPIC FEATURES
1. Case 1 (AFIP 530833)	M 24	RUL	Cough	Chronic cough for 2 months before removal of mass	Tuberculosis	Circumscribed soft spherical mass, 3 cm. diameter. Cut surface, yellow. Central cavity	Fibroblasts, foam cells, plasma cells; hemorrhages. No mitoses
2. Brunn 1939 (AFIP 170825)	F 5½	RUL	Cough Fever	Onset with febrile respiratory infection	Tuberculosis	Circumscribed soft spherical mass, 5 cm. diameter. Cut surface, pink tan	Plasma and other inflammatory cells; fibroblasts; hemorrhages. No mitoses
3. Ford and associates 1950 (AFIP 280217)	F 35	LLL	Chest pain	Repeated attacks of bronchitis	Tuberculosis	Circumscribed spherical mass, 2.5 cm. diameter. Cut surface, yellow	Fibroblasts; hyalinized fibrous connective tissue; foam cells, plasma cells. No mitoses
4. Case 4 (AFIP 485798)	M 41	RLL	Cough	Lobar pneumonia at 19 years; bronchial adenoma removed at 39 yr.	Tumor	Circumscribed firm spherical mass, 7 cm. diameter. Cut surface, yellowish gray	Fibroblasts; collagenous fibrous connective tissue; foam cells, plasma cells; hemorrhages. No mitoses
5. Childress and Adie 1950	M 21	RML	Cough	Chronic cough 8 months	Tumor	Circumscribed firm spherical mass, 4 cm. diameter. Cut surface, yellow gray	Plasma and reticulum cells, eosinophils; dense collagenous connective tissue; foam cells in adjacent alveoli
6. Csernely 1941	M	RUL	?	"Fibroma" of lung removed 7 years previously		Circumscribed firm spherical mass replacing entire lobe	Fibroblasts; hyalinized fibrous connective tissue; foam cells. No mitoses
7. Scott and associates 1948	F 34	RUL	Hemoptysis	Pleurisy 11 years before operation	Tuberculosis	Circumscribed friable spherical mass, 4.5 cm. diameter. Cut surface, yellow	Fibroblasts; hyalinized fibrous connective tissue; foam cells; hemorrhages. No mitoses

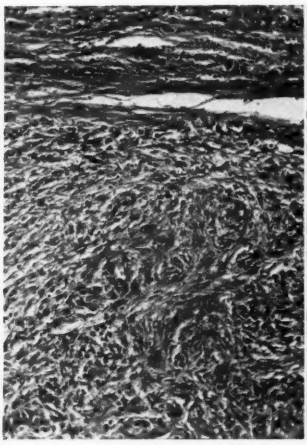


Fig. 13.

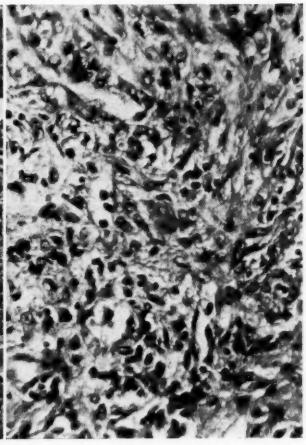


Fig. 14.

Fig. 13.—Case 4: Atypical fibroblasts and inflammatory cells compose the "tumor." (×235.)

Fig. 14.—Case 4: Scattered plasma cells and lymphocytes among connective tissue elements. (×235.)

DISCUSSION

Two of the four cases described have been reported elsewhere,[2, 3] and a search of the medical literature for other examples yielded three additional cases.[4-6] With the increasing frequency of surgical removal of circumscribed pulmonary masses, whether neoplastic of inflammatory, it may be anticipated that these supposedly rare lesions will be discovered in greater numbers.

The clinical and pathologic features of the seven cases are charted in Table I. It is readily seen that all of the lesions were similar in that they tended to have a prolonged course, showed no progressive increase in roentgenographic size, were well circumscribed, and were characterized histologically by fibroblastic and endothelial organization of the diseased area, with varying admixtures of inflammatory cells. Of particular significance were the results of surgical removal, for extirpation of the tumor was completely successful. These tumorlike masses were unrelated to age. The youngest patient was 5½ years and the oldest, 41 years of age. Both sexes were affected. The majority of patients gave a history of respiratory tract infection preceding discovery of the intrathoracic mass. The infection was usually in the form of pneumonia, pleurisy, or recurrent bronchitis. In Cases 2 and 4, the patients had a definite history of pneumonia preceding the appearance of the mass, and in Case 1, a negative x-ray film was recorded four months before the mass was discovered, with a respiratory infection in the interim. Hence it seems justifiable to relate the mass to the antecedent infection.

The presenting symptoms were characteristically mild. Most of the patients had only moderate cough, which was nonproductive or slightly productive of mucopurulent sputum. Only one patient had chest pain. Labora-

tory studies were uniformly within normal limits, and in no instance were pathogenic organisms recovered from sputum or gastric washings.

The roentgenographic appearance of these masses was not diagnostic. Most of the "tumors" were peripherally located, and only one was on the left side. The upper lobe was affected with approximately the same frequency as the lower. The lesions appeared as opaque, sharply circumscribed, spherical masses, with or without central cavitation. The combination of a history of pleurisy, cough, or hemoptysis with the roentgenographic appearance described usually resulted in a clinical diagnosis of pulmonary tuberculosis, and the discovery of "tumor" at operation or autopsy was usually unexpected.

Pathologic Features.—Grossly, all the lesions were sharply circumscribed, spherical nodules from 2.5 to 7.0 cm. in diameter. The degree of encapsulation varied. The consistency of the mass depended upon the relative proportions of its components. Thus lesions which were chiefly xanthomatous were soft and friable; those with a preponderance of plasma cells were moderately firm and granular, and those with dense fibrosis were rubbery. Two of the lesions were distinguished by central cavity formation, but there appeared to be no significant relationship with the bronchi. None of the nodules was directly in contact with pleura.

Microscopically, the most characteristic feature was the combination of several components. These were: (1) spindle-shaped cells; (2) vascular fibrous connective tissue, with or without hyalinizing fibrosis; (3) foam cells with sudanophilic material; and (4) chronic inflammatory cells, chiefly plasmacytes.

The spindle-shaped cells tended to be arranged in interlacing bundles, which, although characteristic of leiomyoma or neurofibroma, were less well organized than those found in muscle or nerve sheath tumors. These cells had abundant pink-staining cytoplasm with vesicular or hyperchromatic nuclei, which often exhibited considerable variation in size and shape. Nevertheless, mitotic figures were absent. These spindle-shaped cells were interpreted as fibroblasts. There appeared to be transition forms between these fibroblasts and the less numerous macrophages. Some of the latter contained brown pigment, which was readily identified as hemosiderin. The amount of collagen appeared to vary with the age of the process. Varying numbers of adult fibrocytes were present. In several instances, the predominance of fibrous components led to an initial diagnosis of fibroma. In one lesion, vascularity was such a prominent feature (Case 1) that the possibility of a vascular tumor had to be excluded.

Foam cells containing sudanophilic material, when present, were often interspersed with fibroblasts, thus giving an appearance of "xanthofibroma" when these cells predominated. Cases 3 and 4 were both reported as "xanthoma." In this group the banal inflammatory cell constituents were less conspicuous, the connective element well defined, and the macrophages with foamy or granular cytoplasm abundant.

Plasma cells were seen in varying number in all the lesions. They were so numerous in Case 2 that the lesion was originally interpreted as a plasma

cell tumor or plasma cell granuloma, although Brunn[2] referred to it as an unusual benign tumor of "contradictory pathology." Lymphocytes and other inflammatory cells were present in smaller numbers, connective tissue elements were fewer, and foam cells were absent.

The lung parenchyma adjacent to the "tumors" usually was not remarkable. Recent hemorrhage, possibly resulting from the surgical procedure, was commonly described immediately around the mass, and areas of fibrosis accompanied by macrophages were sometimes noted.

Diagnostic Considerations.—The clinical features, such as antecedent respiratory infection, long duration without change in roentgenographic size of the mass, absence of recurrence following surgical removal, together with morphologic aspects, including circumscription of the mass, inflammatory features, fibrosis, and lack of mitosis, support the interpretation that these lesions are the result of antecedent infection and are not neoplasms. Although in some instances it was impossible to determine whether an infection actually preceded the tumor, in Cases 1, 2, and 4 the evidence that the fibroblasts and capillaries were part of a hyperplastic repair process, rather than a neoplasm, appeared to be unequivocal.

It is apparent from a review of the literature and a study of the four cases reported herein that these lesions are usually interpreted as neoplasms when a particular cell dominates the histologic picture. This is true of the xanthofibroma reported by Csermely[5] in 1941, and of a similar lesion included in this series. If the fibroblasts or their cellular products form the bulk of the mass, the tumor, of course, resembles a fibroma. Similarly, when plasma cells predominate, the tumor may be classified as a "plasma cell tumor" or "plasma cell granuloma."

The microscopic appearance of the tumors further supports the theory of their inflammatory origin. First, even when one cell type predominates, such as the fibroblast or plasma cell, the cells are sufficiently uniform and regular in appearance that one would hesitate to associate them with neoplasia unless misled by the unusually circumscribed and firm, gross character of the tumor. Moreover, the admixture of other cells, such as lymphocytes and lipoid macrophages, and the free intermingling of all of these histologic elements are other traditional features of chronic inflammation and repair and are not uncommonly found in chronic inflammatory lesions of the lung, such as unresolved pneumonia or an organizing abscess. Finally, fragments of elastic fibers may be demonstrated throughout the inflammatory "tumor," in contrast to their arrangement in true neoplasms in which similar elastic remnants are seen only at the extreme periphery, where replacement of the lung parenchyma is still incomplete.[2]

We are of the opinion that all these "tumors" represent variants of a postinflammatory repair process and are similar to the tumorlike pleural masses reported by Brown and Johnson.[1] Therefore, they may be referred to as inflammatory pseudotumors and are to be carefully differentiated, both clinically and histologically, from true neoplasms, especially sarcomas.[7]

SUMMARY

1. Four postinflammatory "tumors" of the lung are described and compared with three similar ones from the literature.

2. Clinically, the lesions are characterized by a history of infection of the upper or lower respiratory tract and roentgenograms suggestive of tuberculomas or well-circumscribed neoplasms.

3. Morphologically, they closely simulate true tumors, such as xanthoma, xanthofibroma, neurofibroma, fibroma, vascular tumor, or plasma cell tumor.

4. The most important anatomic criteria which should arouse suspicion of postinflammatory "tumors" are: generous numbers of inflammatory cells within the mass, maturity of the fibroblastic components, lack of mitotic activity, and remnants of pre-existing parenchyma throughout the mass in the form of elastic fibers.

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CILIATED EPITHELIAL CYST OF THE ESOPHAGUS ASSOCIATED WITH CARDIAC ABNORMALITIES

Report of Two Cases

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Houston, Tex.

CILIATED epithelial cysts of the mediastinum comprise an interesting group of congenital anomalies which are being encountered with increasing frequency. The commonest type is found in the region of the tracheal bifurcation or main stem bronchi. Because these are thought to be derived from a pinching off of a bud from the primitive trachea, they are called bronchogenic cysts. The second, less common type of ciliated epithelial cyst, which occurs in the wall of or adjacent to the esophagus, has been variously labeled "esophageal," "foregut," or "ciliated epithelial esophageal" cyst.

Characteristically located in the lower third of the esophagus, these cysts usually project to the right. Histologically, they are composed of a lining epithelium of pseudostratified, columnar, ciliated cells resting on a layer of submucous connective tissue, which may contain scattered tubular glandular formations and some smooth muscle. Stratified squamous epithelium may also be present in some areas. Since many of the esophageal cysts are intramural, the wall generally contains, in addition, muscle fibers which are derived from the esophageal muscular coats. Plates of hyaline cartilage are occasionally found within the walls of the cysts. The lumen is filled with a grayish-white viscid fluid resembling boiled starch. There is no communication with the lumen of the esophagus.

The first report of an esophageal cyst is credited to Wyss,[21] who, in 1870, described a tumor in the posterior wall of the esophagus, 3.8 cm. above the gastric cardia; the lining of the cyst was lost. In 1913 Pappenheimer[16] collected from the literature 14 cases of ciliated esophageal cyst and added 2 of his own, and in 1944 Ranstrom[18] reported 17 cases. A recent review of the literature has disclosed 16 other cases,[2, 3, 5, 6, 7, 8, 10, 11, 12, 13, 14, 15, 17, 20, 22] making a total of 49 reported cases.

Reports of cardiac symptoms associated with mediastinal tumors are extremely rare. In 1931 Sauerbruch[20] removed an epithelial cyst from the lower third of the esophagus of a 13-year-old boy who was suffering from dyspnea, cardiospasm, and extreme cardiac irregularity. Following removal of the cyst, these symptoms disappeared. Rawls and Ancona[19] reported a case of malignant lymphoma that appeared as a small nodular mass in the left mediastinum near the pulmonary hilum. The patient had paroxysmal auricular fibrillation, which disappeared after the tumor responded favorably to radiotherapy.

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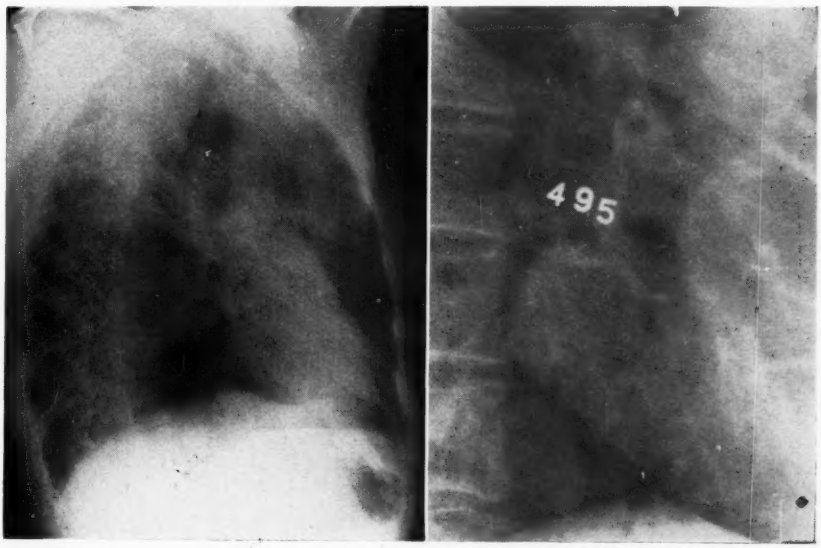
Similarly, Adamson[1] recorded a case of mediastinal tumor associated with sinus arrhythmia, nodal premature beats, and pulsus paradoxus. At necropsy a lymphosarcoma was found attached to the posterior surface of the sternum and involving the posterior wall of the pericardium. Donovan[9] described a case of severe angina pectoris that persisted for eight days and terminated fatally. Necropsy revealed coronary sclerosis with complete occlusion of the right coronary artery and a leiomyoma in the lower third of the esophagus. Because severe thoracic pain of such long duration is uncommon in myocardial infarction, Donovan concluded that the esophageal tumor may have been a contributing factor.

Recently, we operated upon two patients with circumscribed mediastinal tumors lying between the esophagus and heart, both of which proved to be ciliated epithelial esophageal cysts. Both of these individuals had cardiac abnormalities; in one, regression of cardiac symptoms following surgical removal of the cyst suggested a causal relationship. Because of the rarity of such association, the two cases will be described in detail.



A. *B.*

Fig. 1.—Case 1. *A*, Oblique and, *B*, lateral roentgenograms of the chest showing a posterior mediastinal density at the level of the ninth thoracic vertebra.

REPORT OF CASES

CASE 1.—C. D., a 48-year-old white man, was admitted to the hospital on March 8, 1951, for investigation of a mediastinal tumor situated between the esophagus and the heart. A mediastinal shadow was discovered during roentgenologic examination of the upper gastrointestinal tract, which disclosed the presence of a prepyloric ulcer crater.

Physical examination failed to reveal any abnormality except frequent premature cardiac beats. Roentgenographic examination of the chest demonstrated a rounded, sharply circumscribed mass about 6 cm. in diameter along the posterior cardiac border in the region

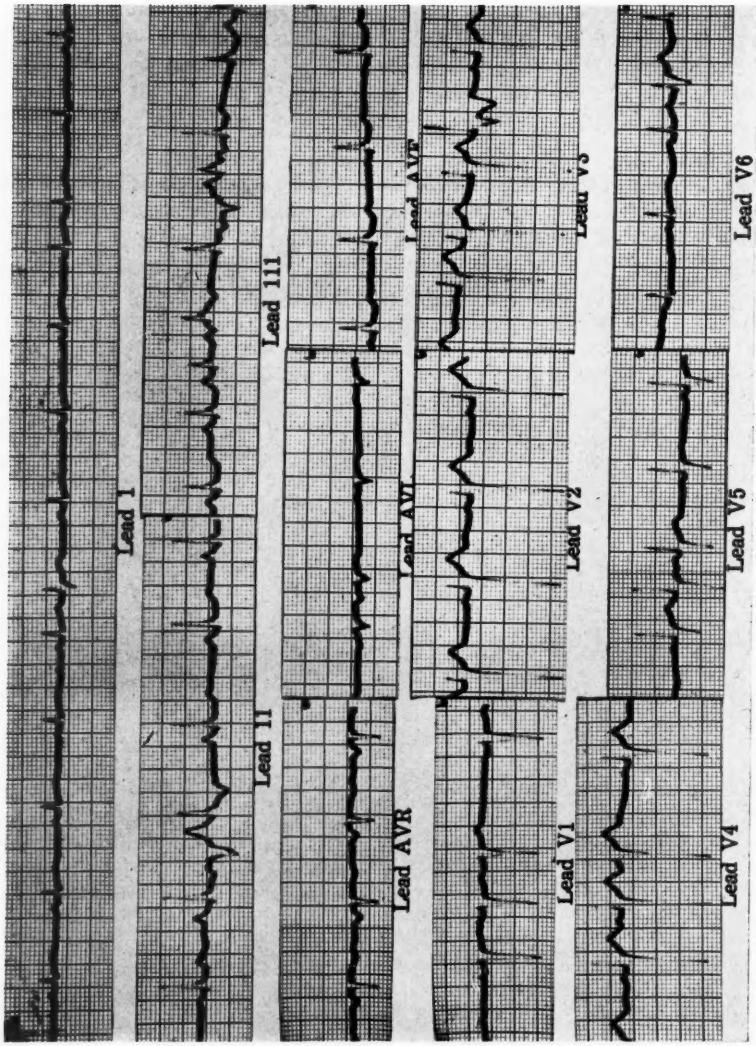


Fig. 2A.—Case 1. Electrocardiograms taken preoperatively, showing auricular and ventricular premature beats.

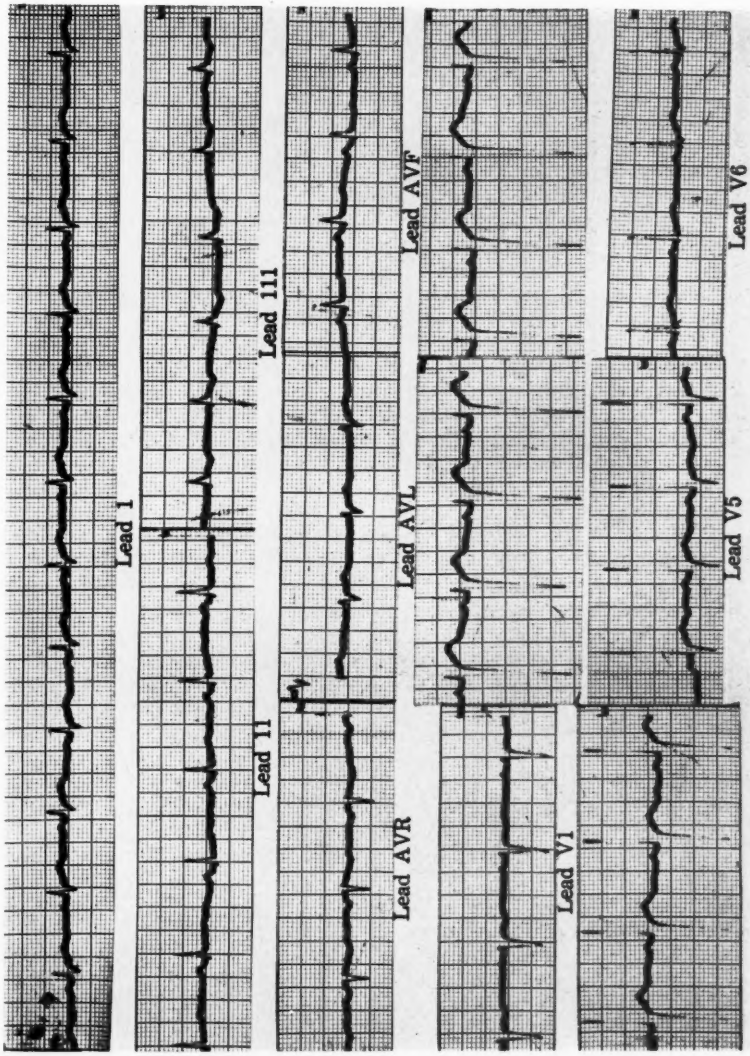
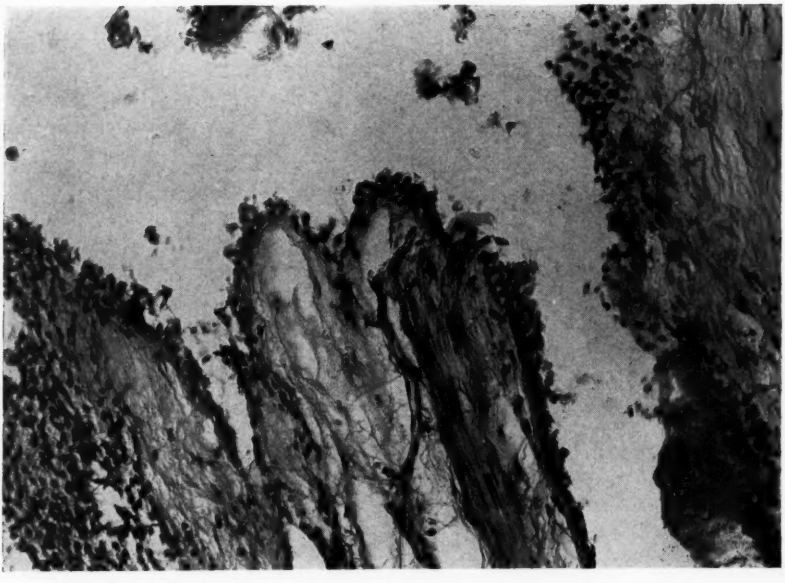


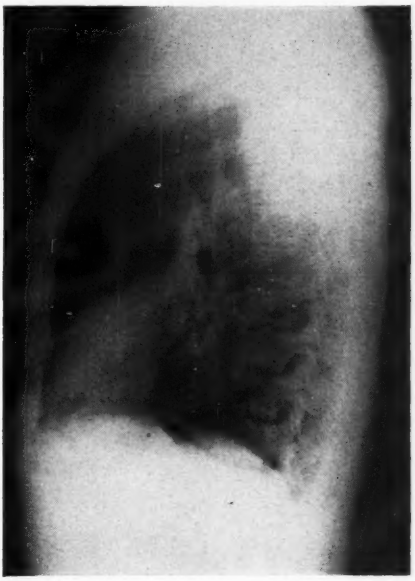
Fig. 2B.—Electrocardiograms taken postoperatively, showing normal rhythm.

A.



B.

Fig. 3.—Case 1. *A*, Photograph of specimen and, *B*, photomicrograph, showing pseudostratified epithelium which is ciliated in places.



A.

B.

Fig. 4.—Case 2. *A*, Lateral roentgenogram of chest, showing posterior mediastinal density at the level of the ninth thoracic vertebra. *B*, Posteroanterior roentgenogram of barium-filled esophagus, showing indentation of left anterior wall by the tumor.

of the right auricle and apparently contiguous to the heart (Fig. 1). On fluoroscopy, the mass moved with cardiac contractions and, with barium in the esophagus, displaced the esophagus to the left, indenting its right anterior wall. An electrocardiogram at that time showed frequent auricular and ventricular premature beats and S-T segments suggestive of myocardial disease (Fig. 2*A*).

On March 13, 1951, exploratory thoracotomy was carried out through a right posterior lateral incision, the chest being entered through the bed of the seventh rib. Retraction of the lung anteriorly revealed a rounded mass within the mediastinum between the heart and esophagus at the level of the ninth thoracic vertebra. The mediastinal pleura over the mass was incised, and, on dissection, the cyst was found to be adherent to the pericardium over

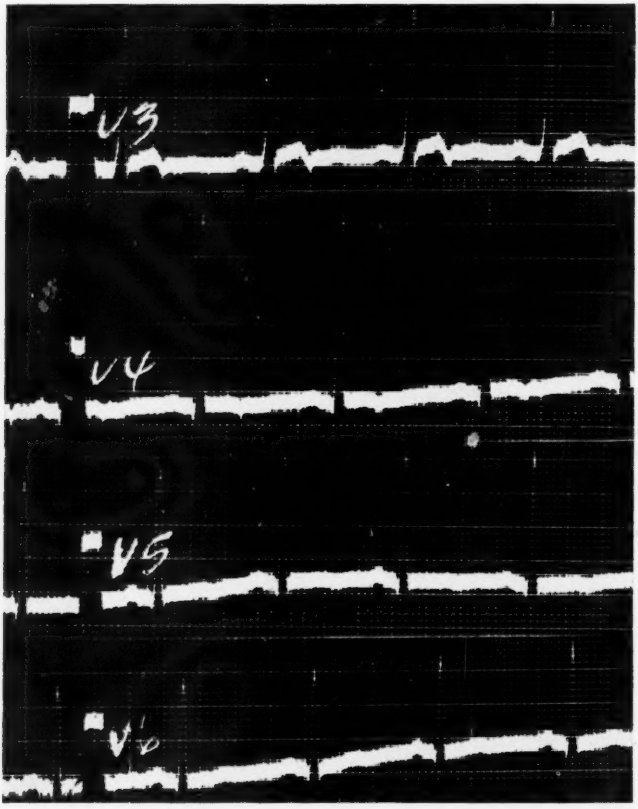


Fig. 5.—Case 2. Electrocardiogram taken before operation, showing T wave changes in Leads V_3, V_4, and V_5.

the right atrium but lying within the esophageal wall. Mobilization of the cyst necessitated opening of the pericardium. The esophageal musculature was incised, and the cyst was removed by blunt and sharp dissection, there being no communication with the lumen of the esophagus. The esophageal musculature was then reapproximated, and the incision was closed.

Postoperatively, the cardiac rhythm became regular (Fig. 2*B*). Convalescence was uneventful, and the patient was discharged on March 20, 1951. When last seen twenty-five months after operation, he had no cardiac symptoms except for occasional episodes of paroxysmal tachycardia.

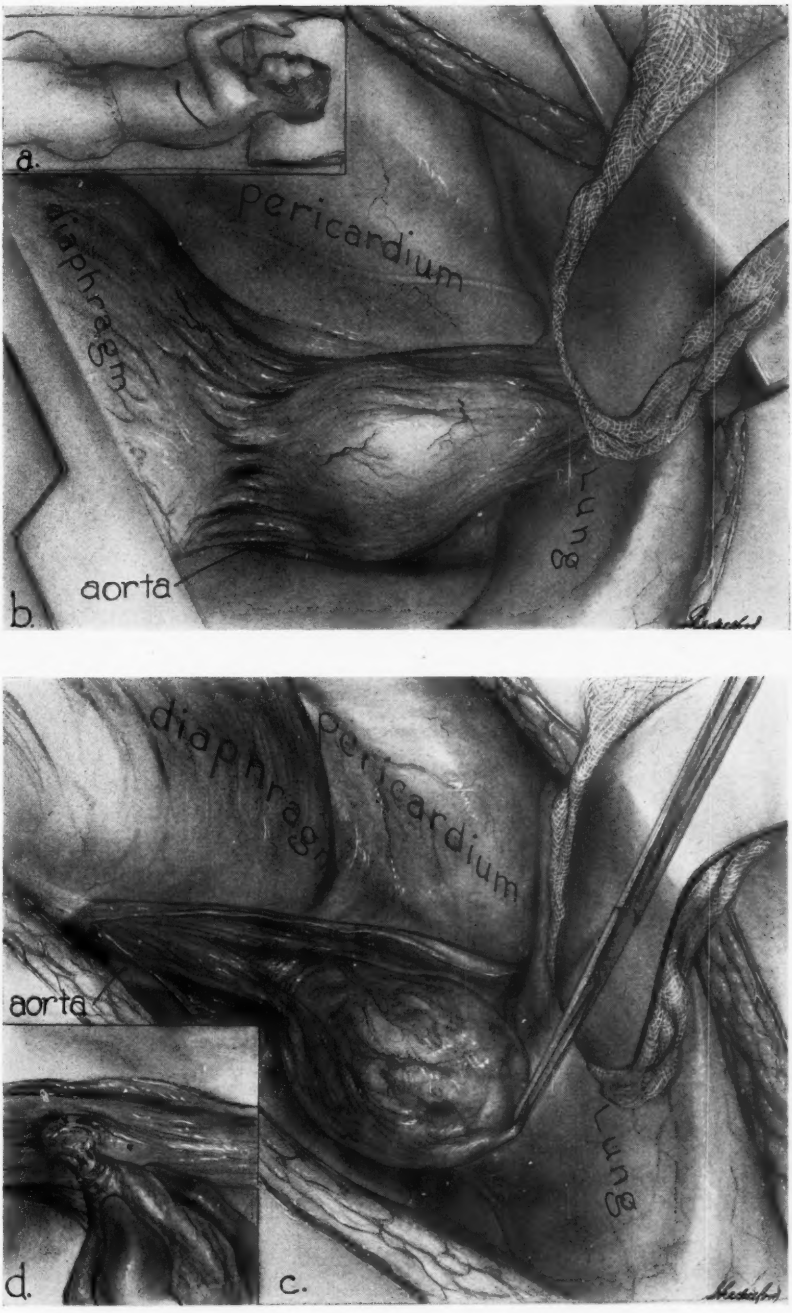


Fig. 6.—Case 2. *a*, The left posterior lateral incision. *b*, Relation of cyst to heart, diaphragm, and pulmonary root. *c*, Extension of longitudinal muscle fibers over cyst indicating its intramural location. *d*, Cyst being separated from submucosa.

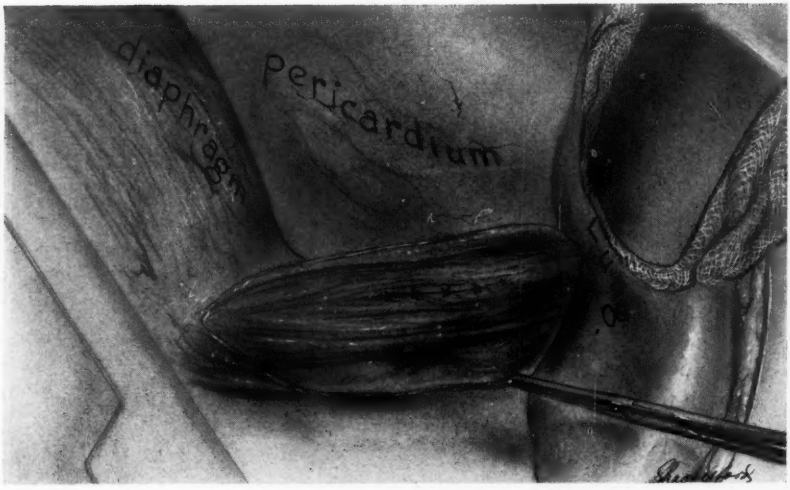
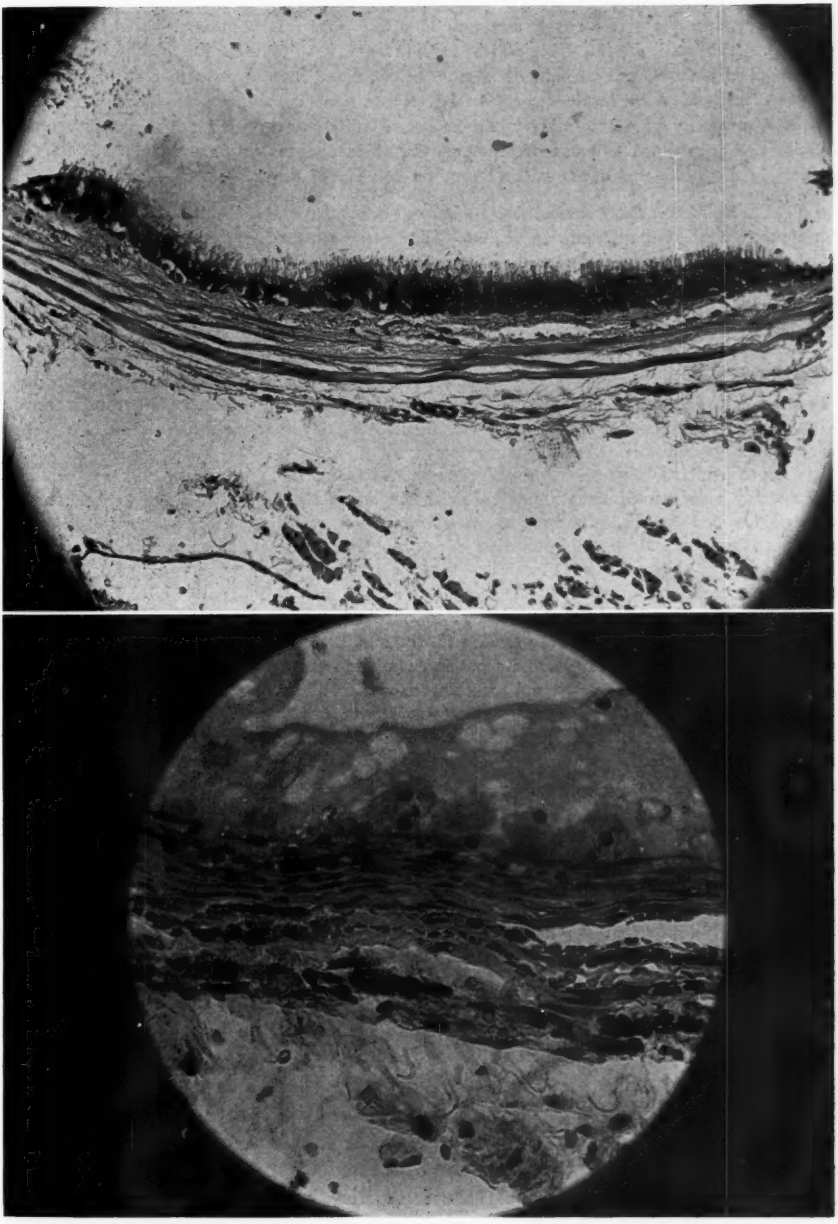


Fig. 7.—Case 2. Reapproximation of esophageal musculature with interrupted silk sutures.

Fig. 8.—Case 2. Photograph of gross specimen.

A.



B.

Fig. 9.—Case 2. Photomicrographs showing *A*, ciliated, pseudostratified lining epithelium, and *B*, hyaline cartilage in wall of cyst.

Pathologic report: Grossly, the cyst was oval and approximately 6 cm. in its long axis. Its external surface was wrinkled and grayish-yellow (Fig. 3, *A*), its lining smooth in parts and rough and papillary in others. It contained viscid, dull-white material. The wall of the cyst, which varied from 1 to 4 mm. in thickness, revealed microscopically a moderate amount of smooth muscle fibers running in a longitudinal and an oblique direction. A moderate amount of edematous stroma was present. The inner lining consisted primarily of a single layer of compressed epithelial cells, although in a few areas definite pseudostratified columnar ciliated epithelial cells could be seen (Fig. 3, *B*). The pathologic diagnosis was developmental cyst of esophageal origin.

CASE 2.—M. D., a 30-year-old white man, was admitted to the hospital on March 4, 1953, for investigation of a retrocardiac mediastinal tumor discovered on routine roentgenography of the chest. Upon close questioning, the patient admitted having rather mild pains in the retrosternal area which radiated to both pulmonary fields.

Physical examination revealed no abnormalities except a Grade II aortic systolic murmur. Roentgenograms of the chest exhibited an oval density lying behind the heart and to the left of the midline at the level of the ninth thoracic vertebra (Fig. 4, *A*). Fluoroscopic examination of the esophagus following ingestion of barium showed indentation of its left anterior wall by the mediastinal tumor (Fig. 4, *B*). An electrocardiogram revealed T-wave changes in Leads V_3, V_4, and V_5 indicative of myocardial disease (Fig. 5). The esophagus appeared normal on esophagoscopy.

A preoperative diagnosis of esophageal cyst was made, and, on April 2, 1953, exploratory thoracotomy was carried out through a posterior lateral incision, the chest being entered through the bed of the left seventh rib. Exploration revealed a cystic mass within the posterior-inferior mediastinum extending from the left inferior pulmonary vein to within 2 to 3 cm. of the cardia (Fig. 6). The pulmonary ligament was divided, and the mediastinal pleura was reflected. The cyst lay within the wall of the esophagus and was intimately attached to the pericardium over the left atrium. By its projection to the left, it was snugly interposed between descending aorta and heart. The cyst was removed, along with the muscle fibers adherent to its outer surface, no communication with the esophageal lumen being found. The esophageal musculature was then reapproximated (Fig. 7), and the thoracic incision was closed. Convalescence was uneventful, and the patient was discharged on April 8, 1953. An electrocardiogram taken three weeks postoperatively indicated improvement in the abnormality previously noted; however, the aortic systolic murmur remained unchanged.

Pathologic report: Grossly, the specimen appeared gray-brown in color, measured 6 by 4 by 4 cm., and had a smooth, glistening external surface (Fig. 8). The wall of the cyst measured 0.1 cm. in thickness, and was of even width; it consisted microscopically of dense hyalinized fibrous connective tissue that was acellular in places. The lining epithelium was composed partially of flat cuboidal cells and partially of pseudostratified ciliated cells. In some sections hyaline cartilage was present in the wall of the cyst (Fig. 9). The lumen was filled with a homogenous gray gelatinous substance. The pathologic diagnosis was ciliated epithelial mediastinal cyst.

DISCUSSION

Several theories have been advanced concerning the genesis of these mediastinal cysts. According to Bremer,[4] gastrointestinal cysts arise as diverticula from the primitive gut during the 20 mm. stage of the embryo. These outpouchings of epithelium may come to lie in the submucosa, between the muscle coats, or outside the gut wall and, when separated from the lumen, become cysts. Others are of the opinion that these anomalies represent congenital tracheoesophageal fistulas that have become occluded anteriorly and posteriorly. Maier[13] has labeled all ciliated epithelial mediastinal cysts as bronchogenic, although he

admits that some of the esophageal cysts may have a separate origin. Ranstrom[18] reported two cases of esophageal cysts, each of which contained a muscularis mucosae beneath the ciliated epithelial lining. Since the trachea has no muscularis mucosae, he concluded that these cysts must have originated in the esophagus.

The similarity in histologic structure of esophageal and bronchogenic cysts indicates a common origin. The fact that both trachea and esophagus arise from a common anlage, the primitive pharynx or tracheogular primordium, suggests that these two types of cysts have at least a common time, if not a common site, of origin. It is probable that, at about the time of separation of trachea and esophagus (4 to 5 mm. stage), tissue fragments are sequestrated which contain either multipotential cells or both entodermal and mesodermal components. At this stage both esophagus and trachea are lined with ciliated epithelium. Fragments near or attached to the trachea remain associated with the tracheobronchial tree and comprise the bronchogenic group of cysts, whereas other sequestrated groups of cells in the wall of or near the esophagus become esophageal cysts. The rapid elongation of the esophagus, which occurs at the time of descent of the heart, accounts for the development of many of the cysts in the lower third of its length.

The anomalies which have been described are not to be confused with the enterogenous or gastrogenous cysts occasionally found in the mediastinum, often attached to the esophagus. These cysts generally contain all the normal structural elements of the stomach or intestinal tract, including a functioning lining epithelium. Bremer[4] referred to these anomalies as duplications rather than diverticula.

Ciliated epithelial cysts, whether bronchogenic or esophageal, appear to be slowly growing tumors which, in the absence of infection, rarely produce symptoms. Most of those reported have been discovered incidentally either at necropsy or during routine roentgenographic examination of the chest. Bronchogenic cysts located about the tracheal bifurcation, particularly in the subcarinal region, may produce severe respiratory symptoms in the child, and cysts in the esophageal wall may cause partial obstruction of that organ.

Even in the absence of symptoms, surgical exploration is indicated for lesions presumed to be mediastinal cysts, since a definite diagnosis is impossible by other means. The treatment of choice is primary excision of the cyst, which has proved both safe and practical. Sauerbruch,[20] in 1931, was perhaps the first to perform primary removal of an esophageal cyst. Prior to that time, the few patients operated upon were treated by marsupialization of the cyst, with removal subsequently. Frequently, both longitudinal and circular muscle layers must be divided in order to expose the cysts, if they lie in the submucosa. Occasionally, the muscle fibers overlying the cyst must be excised if they are adherent. As these lesions do not communicate with the esophageal lumen, they can usually be removed without disturbing the mucosa.

It is interesting to speculate on the possible relationship between the retrocardiac esophageal cysts and the cardiac abnormalities in the two cases

herein reported. In the first (C. D.) the tumor lay behind the right atrium and from this position may have compressed the regions of both the sinoauricular node lying at the junction of the superior vena cava and the right atrium and the auriculoventricular node at the base of the interauricular septum. Conceivably, this pressure from the cyst may have set up aberrant stimuli which resulted in the premature beats. On the other hand, it has been demonstrated that cardiac contractions may be initiated from any section of the heart. The irregularity of cardiac rhythm may have arisen, then, from an area of the auricular and ventricular wall in contact with the cyst and not from the nodal area. It should be pointed out, however, that there is hardly more than suggestive evidence of a causal relationship between this cyst and the cardiac abnormality. The etiology of premature beats is not always apparent, and the cause for their disappearance may be equally obscure, so that a causative role cannot be assigned to any single factor unless the evidence is definite.

In the second case (M. J.), the preoperative electrocardiogram was abnormal, and although postoperative tracings showed some improvement, there is no real evidence that removal of the cyst was responsible for this, since bed rest alone may result in disappearance of this type of electrocardiographic abnormality. Further follow-up studies are needed to establish definitely the relationship of the mediastinal tumor to the cardiac abnormality.

SUMMARY

Ciliated epithelial cysts of the esophagus, though comparatively rare, are being recognized with increasing frequency. Reports of 49 cases have been found in the literature. Even rarer are reports of cardiac symptoms associated with mediastinal tumors. We were able to find only 4 such cases in the literature.

Two cases of ciliated epithelial esophageal cysts with associated cardiac abnormalities treated surgically are presented. The anatomic proximity of the cysts to the heart and the electrocardiographic reversion toward normal, following excision of the cysts, suggest an etiologic role of mediastinal tumors in the production of cardiac abnormalities.

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BILATERAL INTRATHORACIC NEUROFIBROMAS OF THE VAGUS NERVES

With a Note on the Physiologic Effects of Cervicothoracic Vagectomy in Man: A Case Report

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BILATERAL neurofibroma of the vagus nerves, unassociated with generalized neurofibromatosis, appears to be an uncommon condition. Apparently no previous case of this type has been reported, although several authors have noted neurofibromas of the vagus nerves associated with cutaneous neurofibromatosis. Gerbode and Marguiles[1] have recently reported a case of this latter type, and reviewed the literature on this subject.

This present case of multiple bilateral neurofibromas confined to the vagus nerves presented certain questions as to the optimum surgical management at the time of thoracotomy, because of the presence of large, but apparently benign, neurofibromas proximal to the origin of both recurrent laryngeal nerves. In addition, postoperatively, this patient has afforded a unique opportunity for observation and study of the physiologic effects upon the heart and other organs of bilateral high cervicothoracic vagectomy in man.

CASE REPORT

L. A., a 40-year-old unmarried white farmer, was admitted to the University of Minnesota Hospitals on Jan. 22, 1953, because of a mediastinal mass first noted in a survey chest film taken some four months earlier. The patient had no detectable symptoms from this chest lesion, with the possible exception of a moderate cough present for the preceding four or five months. This cough was attributed to his smoking 10 to 40 cigarettes daily for the past eighteen years. There was no family history of tuberculosis, neurofibromatosis, or lymphoblastoma. The patient had not had previous chest x-ray films.

Physical examination disclosed a well-developed, well-nourished, healthy appearing white man with no abnormalities except some hypertrophy of the pharyngeal tonsils. There were no masses palpable in the neck or supraclavicular areas. The patient had had, in the Outpatient Department before admission, an inguinal lymph node biopsy for an apparently enlarged node. Microscopic examination of this tissue was reported as "benign hyperplasia." Admission urinalysis, hemoglobin, white blood count, differential, and erythrocyte sedimentation rate were within normal limits. Bronchoscopy failed to reveal any abnormality. Likewise, the bronchial washings were negative for acid-fast bacilli on smear, for fungus on culture, and for tumor cells on pathologic examination. An x-ray film of the chest (Fig. 1) showed nodular, soft tissue masses projecting laterally from the mediastinum at the level of the aortic arch. Chest fluoroscopy showed these masses to occupy a paratracheal position in the mid-mediastinum, just lateral to, and somewhat anterior to, the trachea. The consensus preoperatively was that this lesion most likely represented a mediastinal lymphoblastoma.

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Fig. 1.—Preoperative chest roentgenogram showing bilateral nodular soft tissue densities projecting laterally from the mediastinum at the level of the aortic arch.

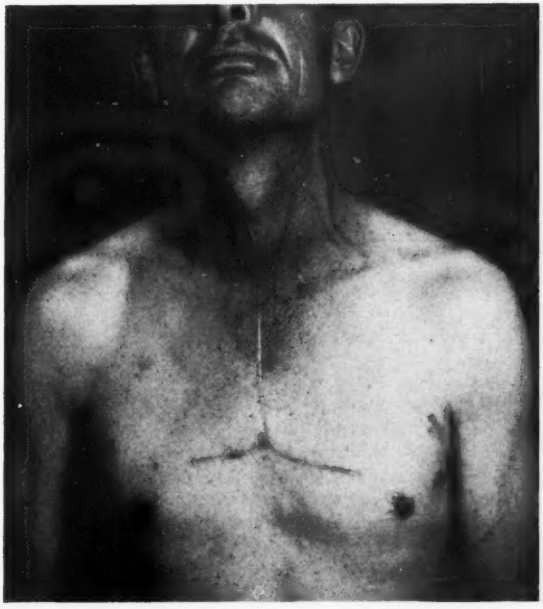

Fig. 2.—Median sternotomy incision with bilateral extensions into the third intercostal space (six weeks postoperatively).

On Jan. 29, 1953, the patient was taken to the operating room, and under Pentothal-curare-nitrous oxide anesthesia, with the patient in the supine position, the chest was opened by a midline sternotomy incision, which was extended bilaterally into the right and left third intercostal spaces. Later in the operation, the incision was extended into the neck in the midline to the level of the hyoid bone (Fig. 2).

The superior mediastinum was then explored. On the left side, a multilobed string of tumor masses was noted to cross anterior to the arch of the aorta. Exploration revealed that the course of these masses was consistent with that of the left vagus nerve. Superiorly, the upper extent of these tumor masses was at the level of the carotid bifurcation. Above that level, the short remaining trunk of the left vagus nerve appeared normal. Inferiorly, the lowermost mass lay behind the pulmonary hilus below the origin of the left recurrent laryngeal nerve. This segment of tumor masses, about 16 cm. in length, was completely removed without difficulty. Exploration of the right side of the mediastinum revealed that the right vagus nerve was afflicted in a similar manner. The

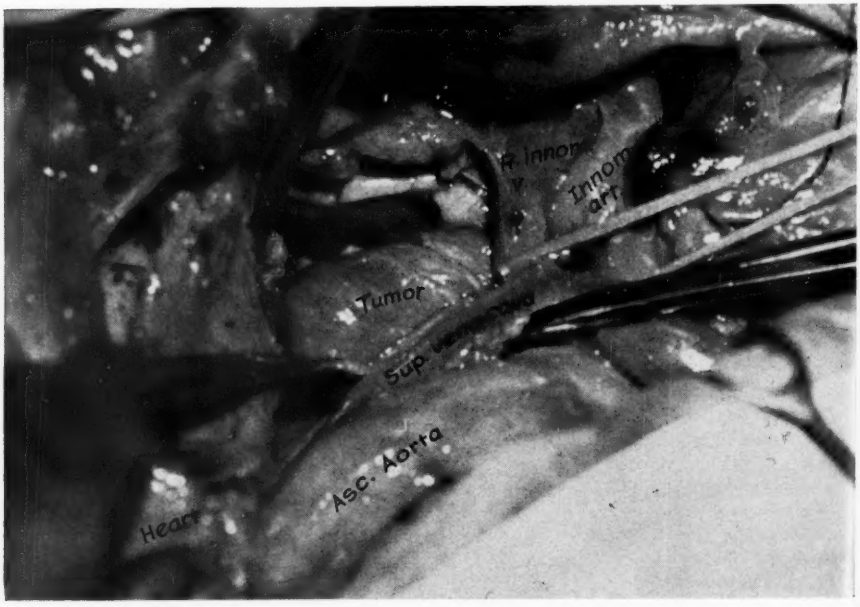

Fig. 3.—Appearance at operation of the neurofibroma of the right vagus nerve located behind the superior vena cava.

lowest mass on this side was the size and shape of a hen's egg, and lay behind the superior vena cava just as it entered the right atrium. Superiorly, this mass terminated in normal-appearing vagus trunk at the level of the right subclavian artery, a few millimeters below the origin of the right recurrent laryngeal nerve (Fig. 3). Just above this short segment of grossly normal vagus trunk and proximal to the origin of the right recurrent nerve, however, another tumor mass involving the cervical vagus trunk was evident. The lower tumor was removed by amputating the vagus trunk superiorly 1 or 2 mm. below the origin of the right recurrent nerve and inferiorly just below the tumor mass. By so preserving the right recurrent laryngeal nerve, it was necessary to leave one remaining neurofibroma undisturbed in the neck on this side.

The patient tolerated the procedure well, except for an abrupt rise in pulse rate from a constant 80 to 110, and a drop in blood pressure from 160 to 140, at the time of severance of the second vagus trunk. After this, the blood pressure slowly rose to 150;

the pulse rose to 120 and remained there for the remaining duration of the surgery. The patient was laryngoscoped just before leaving the operating room; the left vocal cord was noted to be paralyzed, but the right cord moved normally.

Gross examination of the surgical specimens indicated that the tumors varied from small nodules to egg-sized masses (Fig. 4). Between these tumor masses and below the lowest ones, the vagus trunks appeared to be normal. The attached portion of nerve trunk could be traced for some distance into the masses. Gross pathologic examination of the removed tissue showed that the tumor of the left vagus nerve was 16 cm. long, measured 2.5 cm. in its greatest diameter, was encapsulated and lobulated, and was soft in consistency. Microscopic examination disclosed a loose, fibrous structure with small, spindle-shaped cells scattered through a reticular and edematous stroma. A thin, fibrous capsule was present. The tumor of the right vagus nerve was 6 cm. long, and the mass had a similar gross and microscopic appearance to that on the left. The pathologic diagnosis was neurofibromas with no evidence of malignancy.

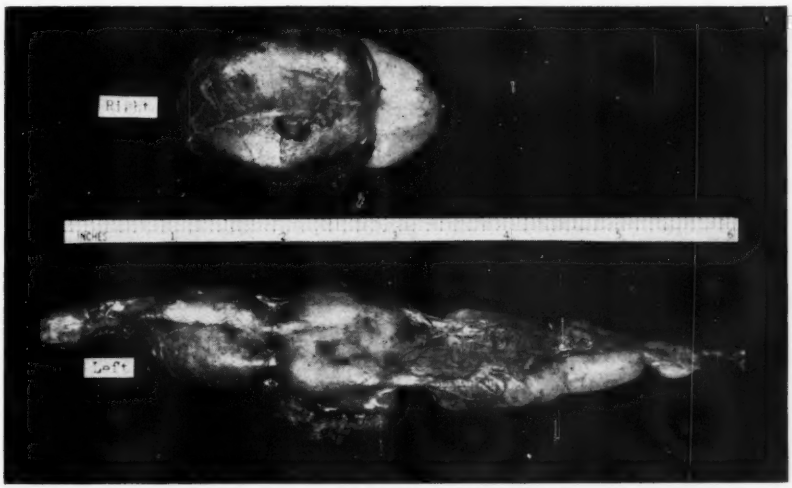

Fig. 4.—Intrathoracic neurofibromas of the right and left vagus nerves removed at surgery. The egg-sized mass lying behind the superior vena cava is shown above. Below may be seen the complete string of neurofibromas involving the left vagus trunk from the level of the carotid bifurcation to the pulmonary hilus.

While the patient was in the recovery room, it was noted that his cough reflex was not present; therefore, the endotracheal tube was allowed to remain in place during that night and was not removed until the following morning. This unusual tolerance to the endotracheal tube was further evidence of a diminished cough reflex, because at that time the patient was fully awake and had no difficulty in voluntarily coughing. About one and one-half days later it became apparent that a tracheotomy would be of value to aid tracheobronchial aspiration of secretions which tended to accumulate because of the poor cough reflex. Thus, tracheotomy was carried out on Feb. 1, 1953. By February 7, it was possible to plug the tracheotomy tube, and the following day it was removed. The remainder of the hospital stay was uneventful, although the pulse rate remained consistently elevated until the time of discharge from the hospital on the thirteenth postoperative day.

The pulse rate of 120 has continued virtually unchanged from the time of its abrupt rise at the severance of the second vagus trunk, through the surgery, during convalescence, and until the time of the patient's most recent visit to the outpatient department twelve months later. Hoarseness, which was marked while the patient was in the hospital, was noted to be considerably improved upon the last visit. Epigastric fullness and "belching,"

which were present for two or three months after surgery, have now disappeared, but a moderate diarrhea (four to six stools each day) still continues one year postoperatively. He is now able to work a full day on the farm, but states that he avoids "heavy" work. Although his neck appears to have some fullness to gross inspection, the tumor mass in association with the cervical trunk of the right vagus nerve is still not palpable one year after surgery. (The fact that there is no palpable enlargement in the right cervical region is probably because of the soft consistency of the tumor mass and its position deep to the sternocleidomastoid muscle.)

DISCUSSION

Nearly all neurogenic tumors of the mediastinum arise from the paravertebral sympathetic nerve trunks and the intercostal nerves; therefore, they are usually found in the posterior mediastinum. In unusual cases, such as in the patient herein reported, they may arise from other nerves and thus may be encountered elsewhere in the mediastinum.

It is of interest in this patient that preoperatively, at a case presentation seminar, an experienced consultant in chest diseases questioned the need for thoracotomy in this patient because the roentgenographic appearance and the location of the masses were considered pathognomonic of lymphoblastoma. It has been our opinion, however, that the relative safety that attends thoracic surgical procedures has outmoded, in most instances, the use of the therapeutic test dose of radiation that was suggested by this consultant as a means of diagnosis. Further, thoracotomy not only permits an accurate diagnosis in such cases, but prevents the occurrence of tissue changes secondary to increasing compression of adjacent organs that may make subsequent surgical removal of even benign lesions more difficult or even impossible. Likewise, in certain benign tumors it is possible that removal will forestall later death caused by malignant transformation.

When it became apparent, at the operating table, that the vagus nerves were involved bilaterally with neurofibromatoma from the level of the carotid bifurcation in the neck to below the origin of the recurrent nerves distally, the decision was made to remove the tumors completely on the left side (where the cervical masses were larger than on the right side), thus sacrificing the left recurrent laryngeal nerve, and to remove all the tumor in the right mediastinum (where the masses were larger than on the left side) up to the origin of the right recurrent nerve. This necessitated leaving at least one tumor mass in the right side of the neck, the lower pole of which was located at the level of the right subclavian artery. Several considerations entered into this decision. To remove completely all of the tumors would have necessitated bilateral sacrifice of the recurrent laryngeal nerves, which is incapacitating. Several consequences make bilateral loss of the recurrent laryngeal nerves a far more crippling procedure than unilateral removal. First, there is loss of speech; second, physical activity is severely curtailed due to inability to increase ventilatory volume because of obstruction to air flow by the paralyzed cords; and finally, there is the obnoxious "crowing snore" which is regularly present during sleep. Other considerations were that the tumors were probably benign, and that one recurrent nerve could be spared, and yet all the intrathoracic tumors could be removed leaving only the one neurofibroma

remaining in the neck where it could be followed by palpation and attended to surgically with relative ease if the need should arise later.

PHYSIOLOGIC CONSIDERATIONS

It is likely that a fairly complete interruption of the parasympathetic innervation to the heart and lungs (as well as to the abdominal viscera) was achieved in this patient; therefore, it seemed pertinent to assess some of the effects of this procedure in this patient.

Bilateral section of the vagus nerves in the neck is said to be uniformly fatal in laboratory animals because of its effect on the cardiorespiratory organs.[2] Markowitz[3] stated, however, that in the dog, if the right vagus nerve is sectioned below the origin of the recurrent laryngeal nerve, the left vagus nerve may be sectioned in the neck without fatal outcome. The present case confirms in man this latter observation.

The difficulty that our patient had with retained secretions (requiring tracheotomy), however, suggests that the protective reflexes of the respiratory tract are seriously, but probably temporarily, impaired by vagectomy at this level. There are several possible explanations for these observations. Action of the cilia is said to be independent of nervous control, but the efficiency of ciliary action is dependent in a large part on the character of the mucous secretions of the respiratory epithelium. It is quite possible that the secretions are adversely influenced by vagotomy, as also may be bronchial peristalsis.[4-6] It is also likely that paralysis of one recurrent laryngeal nerve, with the associated vocal cord paralysis, seriously interferes with the efficiency of the cough reflex because of the virtual impossibility of maintaining a closed glottis against which to expire forcibly, and then suddenly open, as occurs normally in the act of coughing. The observation by Kuntz[7] that the cough reflex may be elicited by stimulation of the vagus fibers distributed to any part of the respiratory tree and the experiments of Cromer and associates,[8] wherein it was noted that bilateral section of the vagus nerves markedly reduced the cough reflex which was manifested following the insufflation of ammonia vapor into the trachea of experimental animals, emphasize additional reasons for this patient's diminished cough reflex.

In view of the extent of the vagus resection and the known vagus pathways to the heart,[9] it is likely that the heart was virtually completely deprived of its parasympathetic innervation while its sympathetic innervation remained intact. The persistently fast pulse rate, still present one year after surgery, and a failure of atropine (1/100 grain) to increase this rate add further support to this conclusion. Moreover, it is a finding of particular interest that in this patient with extensive cervicothoracic vagectomy, both the systolic and diastolic blood pressures have remained for twelve months now persistently elevated by about 20 mm. Hg above their preoperative levels.

Because of the unusual opportunity offered by this case for study of the effect of vagectomy upon cardiac function, Dr. Fraser and Dr. Chapman* have studied this patient's cardiac rate, blood pressure, and cardiac output in response to exercise four months after surgery. The results of these observa-

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tions are reported in detail elsewhere,[10] but in summary several interesting observations were made, as follows: in the resting state the patient had a persistently high pulse rate (120/min.); this rate increased in response to exercise. The magnitude of this increase in pulse rate, however, was distinctly less than normal. On the other hand, the responses to exercise of the cardiac output, blood pressure, and circulation time were all within previously observed normal limits for the work load used.

These observations add further evidence that man has nonvagal mechanisms for increasing cardiac rate and cardiac output in response to exercise. The fact that the magnitude of the increase of the cardiac output in response to graded exercise was normal in this patient, whereas the pulse rate increase was less than normal, indicates that readjustment was brought about in part by an increased stroke volume. These studies also complement those of Smithwick and associates,[11] who found that sympathetic denervation of the heart in man uniformly resulted in a slowing of the heart rate and a reduction of the pulse rate increase in response to exercise.

SUMMARY

A case of bilateral cervicothoracic neurofibromas of the vagus nerves without cutaneous neurofibromatosis or evidence of neurofibromas elsewhere in a 40-year-old man is presented. The surgery performed in removing these tumors involved an extensive cervicothoracic vagectomy, intentionally sacrificing the left recurrent laryngeal nerve but preserving this nerve on the right side.

Postoperatively, some physiologic observations have been made upon the effects in this patient of cervicothoracic vagectomy upon cardiorespiratory function. These alterations in function consisted of a diminished cough reflex, persistently elevated pulse rate and blood pressure, a normal cardiac output with a normal response to exercise, but a diminished pulse rate response to exercise, and a lack of any detectable response of this elevated resting pulse rate to a therapeutic dose of atropine. Some of the physiologic implications of these observations are briefly discussed.

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A METHOD FOR MEASURING TIDAL AND MINUTE VOLUMES IN PATIENTS UNDER REGIONAL AND GENERAL ANESTHESIA

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THE determination of tidal and minute volumes in the anesthetized patient requires the introduction of the measuring meter between the circle-flow system of the gas anesthetic machine and the breathing mask or intratracheal tube. This is necessary because the volume of gas flowing in the afferent and efferent limbs of the circle system of the anesthetic machine bears no fixed relation to the volume moved in respiration of the patient. This same situation pertains in "to and fro" systems to a somewhat less degree, but still of sufficient magnitude to require special arrangement of the metering.

The major reason for this is that the concentrations of the various gases, such as N_2O_2 and O_2, in the anesthetic mixture, are maintained by allowing wide margins of excess flow of these gases to enter the system at constant rates, with the excess escaping at the machine end of the exhalation side of the anesthetic tubing. Other spill-over methods are also used, but all allow the flow of sizable quantities of gases, which have no participation in the respiration of the patient, through the anesthetic machine.

The second requirement for simple, direct metering is the division of the airway into inspiratory and expiratory channels without the interposition of any dead air space or resistance to flow. This allows metering to be accomplished by the measurement of a unidirectional gas flow.

The introduction of a properly valved circle system between the orifice of the gas machine tubing and the patient's mask or intratracheal tube will accomplish the division of the flow into inspiratory and expiratory channels. These channels are unaffected by the flow or pressures occurring in the anesthetic machine or its tubing. The flow in this system will be entirely dependent upon the volume of gas entering and leaving the patient's airway. (See Fig. 1.)

This schematic diagram has purposely simplified the gas machine, omitting its valving, soda-lime, bag, inflow channels, and variable pressure escape port. The two channels interposed between the gas machine and the intratracheal tube are valved so that as the pressure on the gas machine side rises above the intratracheal pressure, the valve on the inspiratory limb opens, whereas the valve on the expiratory limb closes, allowing gas to flow from the machine through the inspiratory side into the lungs. A reversal in pressure relations closes the inspiratory valve and opens the expiratory valve to allow gas flow

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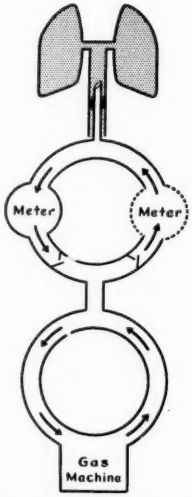

Fig. 1.—A simplified schematic drawing of a closed anesthesia circuit with the gas meter in either the inspiratory or expiratory arm. The meter records only the actual volume of gas that the patient inspires or expires, regardless of any leak in the gas machine.

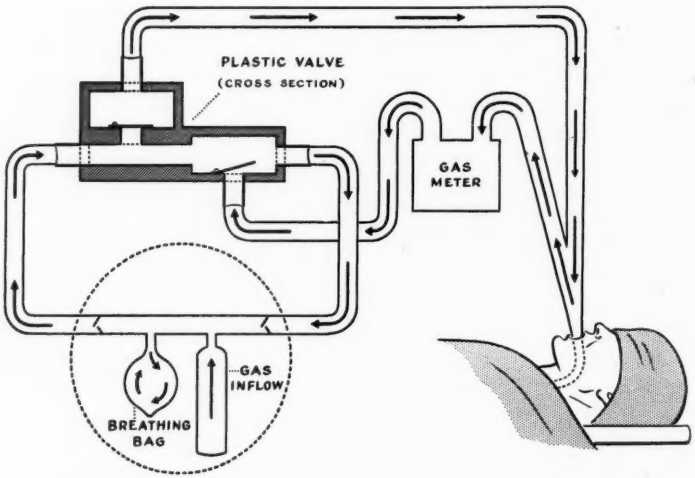

Fig. 2.—Circuit including gas machine, valve, gas meter, and patient. The encircled area schematically represents any standard gas anesthesia machine. The valve positions indicate the expiratory phase of respiration.

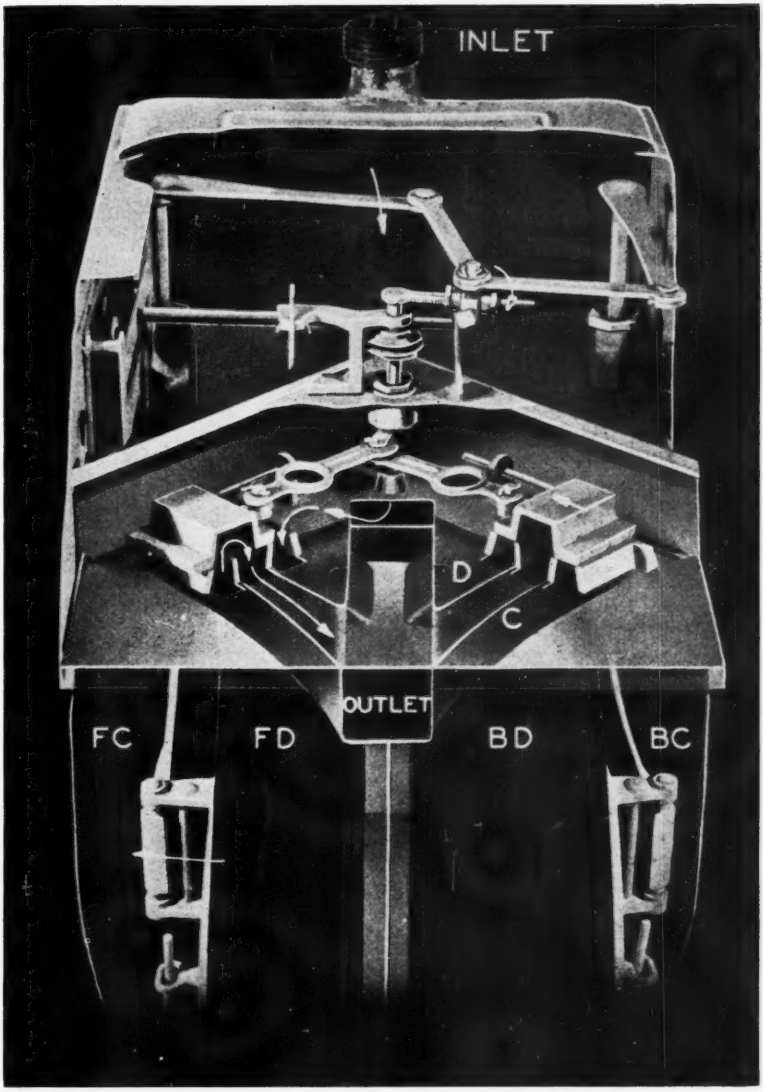

Fig. 3.—A section of the volume displacement meter.[1]

FC—front case compartment
FD—front diaphragm compartment
BD—back diaphragm compartment
BC—back case compartment
D—port to diaphragm compartment
C—port to case compartment

When the valves are in this position, *BD* is filled and *BC* is emptied. Gas entering through the inlet passes through the port of the front diaphragm compartment, filling it. At the same time, *FC* is being emptied, gas leaving through the port of the front case compartment and meter outlet. The front disk is moving away from the center partition, moving the front flag arms toward the center of the meter, rotating the tangent arm and crank in a clockwise direction, and moving the back valve in the direction of the arrow, thus opening the back ports so that gas can enter *BC* and leave *BD*.

through the expiratory limb. Because there is always a slight pressure gradient in one direction or the other across this system, one of the valves is always closed, and circling is avoided.

Fig. 2 is a schematic drawing designed to show the valving. It again greatly simplifies the gas machine and is inaccurate in depicting the intratracheal tube connection. The channels leading into and out of the broken-line circle represent the gas machine tubing. Note that they communicate through the valve block without any valving. The valves are arranged to allow flow out of one tube to the patient when the pressure in the trachea is less than in the machine. The flow is through another tube when the intratracheal pressure is higher than in the machine system. The diagram depicts the inflow and outflow tube joining before meeting the intratracheal tube. This is in error, as it would materially increase dead space. In actual application, these tubes are standard anesthetic tubing, or one-inch flexible but rigid smooth-bore plastic tubes attached to a standard intratracheal fitting or mask fitting. The meter is shown here in the expiratory side, which is preferable. The valve block is of clear plastic, allowing visualization of valve action at all times.* The valves are rubber-faced gravity type made of brass disks hinged at one point. The block should be made so that it can be disassembled for servicing and cleaning, but it must be airtight.

METERING

We have found that the ordinary volume displacement meter (manufactured by the American Meter Company) of the type used by most commercial gas companies for dwelling installation is very satisfactory. The American Meter Company rates these meters as being accurate within 0.25 per cent and offering resistance of 0.25 cm. H_2O at 9.44 liters per minute. Our measurements show them to be accurate within 1 per cent and to offer less than 1 cm. H_2O resistance at flows less than 60 liters per minute. These meters are encased in opaque metal boxes, and a description of the structure of such a meter may be found in the literature.[1]

These meters record in cubic feet. For convenience a long hand was substituted for the conventional index on the one-half cubic foot scale, and a large dial calibrated in liters was placed behind this hand. For minute volume studies a stop watch must be used with the meter for timing. Tidal volumes may be read directly from the liter scale.

The meter may be placed in either the inspiratory limb of the circuit or the expiratory side. The latter is preferable if positive pressure assisted or controlled respiration is carried out, as any leakage of the mask or about the intratracheal cuff will occur during the positive pressure inspiratory phase. Stead and associates[2] subsequently adopted this system of divided airway and metering in studies on patients undergoing thoracic surgery. They found no appreciable difference in minute volume readings from placing the meter in either the inspiratory or expiratory arm of the circuit with a tight-fitting intratracheal tube.

For evaluation of this system and as a base line for further studies, normal subjects under local or spinal anesthesia were studied. These patients were all fully conscious, had no airway difficulties, and experienced no sense of dyspnea or showed any increase in respiratory rate or effort while breathing through the equipment by means of a tightly fitting face mask.

*A sticking valve allows to-and-fro motion, producing a large dead space of real hazard to the patient.

A gas machine was not used in these subjects, but rather the ports on the valve block, where the gas machine tubing attaches, were left open to exchange room air. Minute volume in these subjects ranged between 5.02 liters per minute to 8.22 liters per minute, varying with the size of the subject, his pain, and anxiety.

A study of normal subjects under general anesthesia having abdominal or extremity surgery, using either a tightly fitting face mask or a cuffed intratracheal tube, showed minute volumes ranging from 8.22 liters per minute to the woefully inadequate figure of 0.85 liters per minute, depending upon the depth of anesthesia and efficiency of respiratory assistance provided by the anesthetist.

CONCLUSIONS

1. Accurate tidal and minute volumes may be determined by providing a double-channel airway properly valved to avoid to-and-fro dead space and by separating the flow into an inspiratory channel and an expiratory channel. The respiratory airway may be divided into inspiratory and expiratory channels by proper valving of a double-channel airway without increasing to-and-fro dead space.

2. This system may be attached to a tightly fitting face mask or cuffed intratracheal tube and the flow measured in either channel by an accurate, low impedance meter for the determination of tidal and minute volumes in conscious or unconscious subjects.

3. This system may be interposed between the outlet of the usual gas machine tubing of either circle or to-and-fro systems without disturbing the gas flows occurring in the gas machine or its tubing, without interfering in any way with the anesthetist's management of respiration, and without recording any volumes except those actually exchanged by the patient.

4. An ordinary volume displacement meter of the type usually used by commercial public service gas companies for dwelling installations, manufactured by the American Meter Company, is admirably suited for measurements of this nature, being sufficiently accurate and having very low flow resistance.

5. Minute volumes under low spinal anesthesia and regional anesthesia are well maintained in the normal physiologic range, whereas with general anesthesia they frequently fall to dangerously low levels.

6. Continuous measurement of minute volumes is essential for the interpretation of data relating to the oxygenation and carbon dioxide elimination of subjects under general anesthesia.

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STAGED AND SIMULTANEOUS BILATERAL PULMONARY RESECTION FOR TUBERCULOSIS

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THE frequency of bilateral pulmonary tuberculosis and the development of pulmonary resection as a form of surgical treatment provoke consideration of bilateral resection. The bilateral approach can be carried out either in stages or simultaneously. The experience to be reviewed in this report is comprised of twelve staged and three simultaneous procedures. Fifteen patients underwent a total of twenty-seven operations without any deaths.

Pulmonary resection for tuberculosis since the first successful planned lobectomy in 1934 has enjoyed an increasing and deserved popularity.[1, 2] Reports since the advent of antimicrobial preparation have been encouraging.[3,4] The incidence of bilateral surgery is not as high as one might expect. Chamberlain reported a series of 300 segmental resections with ten bilateral cases.[5] Kelley listed nine bilateral resections in 104 cases.[6] A possible explanation is that the contralateral lesion may regress in the interim between the initial and second operations. This has been our experience in a number of instances where prior to the first operation the period of repetitive x-ray observation has been relatively brief. Staged bilateral resection, nevertheless, has a definite place in the surgical treatment of pulmonary tuberculosis. The idea of simultaneous bilateral resection is attractive because it offers a single operation rather than two with a substantial time interval. Another point in favor of the simultaneous program is the obligatory participation of all residual segments in the respiratory effort. This may provide a more efficient respiratory apparatus than is available to the staged patient whose first side doubtless carries the major share of respiratory function. Where pulmonary capacity has been reduced by initial surgery, respiratory function may be critical. Cyanosis has been noted following completion of the staged procedures. One patient (Patient 9 of the staged group) required tracheotomy to decrease the dead space and to insure lung capacity against atelectasis. There was no cyanosis in any of the simultaneous bilateral segmental resections.

Possibly the first simultaneous bilateral thoracotomy was performed by Block of Danzig in 1882.[7] The patient was a young woman who requested the operation for her suspected tuberculosis. Normal lung was found at operation, which may have persuaded Block to open the contralateral chest. It is not clear whether any resection was performed; however, no disease was found.

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The patient expired either on the operating table or very early in the postoperative period. Legal proceedings were instituted, and Block took his own life.[8] The actual circumstances surrounding this clinical misadventure are somewhat equivocal. Block as well as Theodor Gluck of Berlin published experimental pulmonary resections on a variety of animals including dogs, pigs, rabbits, horses, and even cows.[9-11] No simultaneous bilateral resections were performed experimentally. The technique consisted of exteriorizing a pulmonary lobe through a small intercostal incision, applying a tourniquet-ligature about the secondary hilus, and excising the lobe. A clinical case had been similarly treated by Kirchhoff in 1876.[12]

Much has been said concerning bilateral resections for bronchiectasis.[13-16] Overholt in 1952 reported the first simultaneous bilateral pulmonary segmental resection for bronchiectasis.[17] An inspissated tuberculous cavity was removed by wedge excision from the posterior segment of the left upper lobe in one patient; otherwise the three cases described were unrelated to pulmonary tuberculosis.

SELECTION OF PATIENTS

Our experience corroborates the proposition that tuberculosis usually inhabits the apicoposterior segment of the left upper lobe, the apical and posterior segments of the right upper lobe, and the superior segments of either lower lobe. This does not mean that segmental boundaries are respected by the tuberculous process, but in general, the site is predictable from x-ray films. Here and there an adjacent subsegment must be sacrificed to excise the principal disease. This is readily done when indicated. In spite of this remarkable constancy of position, one must adapt himself to the disease encountered and not perform a segmental resection for tuberculosis which is of lobar or greater dimension. Planigrams in particular are helpful in avoiding this pitfall. Bronchoscopy also is valuable since the patient with endobronchial disease must have either a longer period of chemotherapy of a more extensive operation.

All cases in either group are of patients at the Anoka State Mental Hospital and, therefore, carry the additional problem of severe mental illness. Bed rest is an impossible discipline, and incidentally, the consequences of its absence have not been striking. A patient is chosen for bilateral resection who over a period of time, usually one or two years, has been unable to secure x-ray clearance for return to the general population of mental patients. Medical treatment has been unable to eradicate the disease, which is well localized to the upper lung fields in most cases. There must be direct bacteriologic evidence of tuberculosis in the form of culture or guinea pig inoculation. Antimicrobial drugs are now given for at least three months prior to surgery.

If clinical findings, x-ray investigation, and bacteriologic history indicate that a patient is definitely a candidate for bilateral surgery, the next question is whether the plan should be staged or simultaneous. Here, physical condition becomes the supreme consideration. One cannot expect a robust subject

when he is dealing with a patient who requires bilateral operations, but only the best risk group should be relegated to the simultaneous plan. Our simultaneous bilateral experience at this time has been at the segmental level; nevertheless, it is conceivable that lobectomy either paired or in combination with segmental resection could be carried out if clinical conditions warranted. Seldom, if ever, should the surgeon reach a situation where the resection on one side has become so formidable that the second side must be abandoned. Careful roentgenographic work-up should prevent this occurrence. Simultaneous bilateral resection is feasible in that patient who requires bilateral surgery and whose general condition is reasonably good.

TECHNICAL CONSIDERATIONS IN SIMULTANEOUS BILATERAL RESECTIONS

The supine position is used with a sandbag along the dorsal vertebral column to elevate the mediastinum. A transverse incision is made in the third anterior intercostal space cutting across the sternum. This provides excellent exposure of either hilus and permits the use of two operating teams at least until both lungs are freed of adhesions and hilar dissection begun. At this point the operating table is rotated for better access to the hilus under consideration. The indicated procedure is carried out on the more involved side first with the contralateral resection to follow. Total operating time is little more than for the customary unilateral segmental resection. No real problem is occasioned by the bilateral pleural opening. Experiences with other forms of chest surgery have demonstrated the relative ease with which this situation is tolerated.[18] Postoperatively, it is advisable to attach the chest catheters on either side to separate suction systems. Adapters and Y tubes connecting the chest catheters to suction should be made of unbreakable material. There probably ceases to be a communication between the two chest cavities after twenty-four hours.

A Pentothal-curare mixture is employed for induction and endotracheal intubation with subsequent addition of nitrous oxide and oxygen in a 1:1 ratio. This permits use of the electrocautery unit in the early stages of the procedure. Since cyclopropane interferes little with circulatory homeostasis and allows delivery of a 90 per cent oxygen content in the gas mixture, this agent is used for maintenance as soon as the cautery is dispensable. The lungs are inflated to the extent that there is no encroachment upon the surgical field. Respirations are either assisted or controlled thoughout. Slight Trendelenburg position is helpful in neutralizing the loss of negative intrapleural pressure and aids venous return to the right heart. This position also facilitates the removal of secretions by the endotracheal suction catheter.

DISCUSSION OF EARLY RESULTS

Since the first of the staged procedures was completed little more than one year ago, it is much too early to speak in terms of end results. The total experience is summarized in Tables I and II. Criteria for discharge from the tuberculosis section must be rigid because of the congested living area to which mental patients are necessarily returned. Clinical and x-ray clearance plus

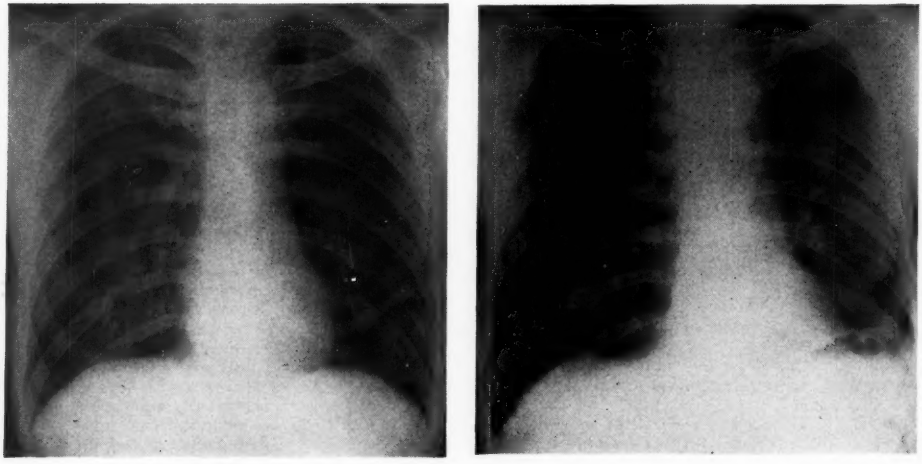
TABLE I. GROUP I, STAGED BILATERAL RESECTIONS

PATIENT	AGE SEX	DATE 1ST OP.	1ST OP.	DATE 2ND OP.	2ND OP.	REMARKS
1	28 F	1-16-52	Ap. and p. segs. RUL	1-19-53	Ap. p. and ant. segs. LUL, sup. seg. LLL, 3-rib thoracoplasty	No complications; x-ray, clinical, and gastric clearance
2	49 M	2-6-52	Ap. p. seg. LUL, 4-rib thoracoplasty	6-22-53	Ap. and p. segs. RUL, wedge sup. seg. RLL	Wound dehiscence after 2nd operation
3	40 M	6-18-52	Ap. p. seg. LUL, wedge sup. seg. LLL	12-3-52	Ap. and p. segs. RUL, wedge sup. seg. RLL	Gastric relapse
4	50 M	6-25-52	Ap. p. seg. LUL	1-5-53	Wedge ap. seg. RUL	No complications; x-ray, clinical, and gastric clearance
5	54 M	7-7-52	RULobectomy, 4-rib thoracoplasty	11-17-52	Ap. p. seg. LUL	No complications; x-ray, clinical, and gastric clearance
6	50 M	7-16-52	Ap. and p. segs. RUL	11-2-53	Ap. p. seg. LUL	Post-2nd-op. air leak eventually resorbed
7	57 F	8-4-52	RULobectomy	1-26-53	Ap. p. seg. LUL	No complications; x-ray, clinical, and gastric clearance
8	55 F	1-14-53	Ap. and p. segs. RUL, wedge sup. seg. RLL	5-20-53	Ap. p. seg. LUL	No complications; x-ray, clinical, and gastric clearance
9	50 F	1-25-53	Ap. p. seg. LUL	5-25-53	P. seg. RUL, wedge ant. and ap. segs. RUL	Post-2nd-op. cyanosis; tracheotomy
10	45 M	2-11-53	Ap. and p. segs. RUL	10-26-53	Ap. p. seg. LUL	No complications
11	63 M	3-4-53	Ap. and p. segs. RUL	12-23-53	Ap. p. seg. LUL	No complications
12	62 M	3-11-53	Ap. p. seg. LUL	12-7-53	Ap. and p. segs. RUL	No complications

TABLE II. GROUP II, SIMULTANEOUS BILATERAL SEGMENTAL RESECTION

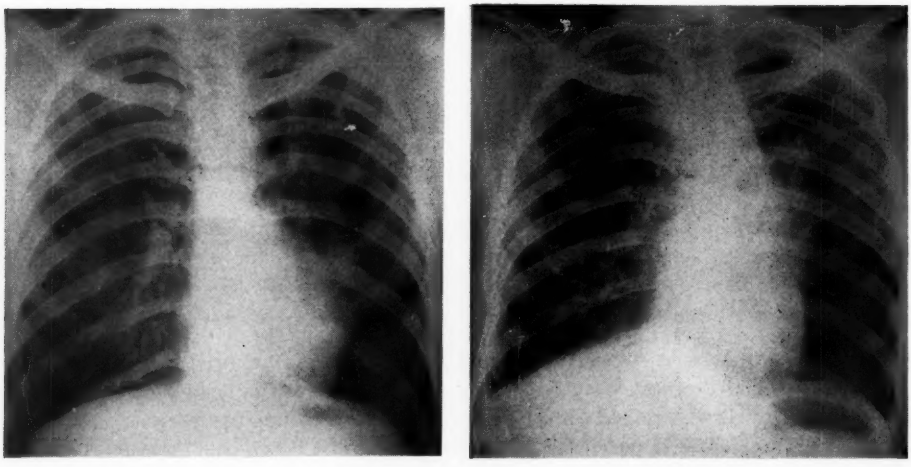
PATIENT	AGE SEX	DATE OF OP.	MORE INVOLVED	CONTRALATERAL	REMARKS
1	23 M	1-6-54	Ap. p. seg. LUL, wedge lingula	Ap. and p. segs. RUL	No complications
2	42 M	1-20-54	Ap. and p. segs. RUL	Ap. p. seg. LUL	Air leak on right; responded to increased suction
3	35 M	2-3-54	Ap. and p. segs. RUL	Ap. p. seg. LUL	No complications

six negative gastric lavage cultures represents a reasonably strict program. Antimicrobial therapy is continued for six months into the postoperative convalescence. In the staged group, drugs are given continuously between operations. One month is permitted to elapse after drugs before the gastric lavage series is initiated. Typical preoperation and postoperative chest films in both the staged and simultaneous groups are seen in Figs. 1 and 2. To this date one case has developed evidence of persistent disease (Patient 3 of the staged group). The postoperative work-up is complete on five other patients, and fractional data are encouraging on the remainder. The simultaneous bilateral



A. *B.*

Fig. 1.—Patient 10 of the staged group: *A*, preoperative roentgenogram; *B*, two months following completion of bilateral resection.



A. *B.*

Fig. 2.—Patient 1 of the simultaneous group: *A*, preoperative roentgenogram; *B*, two months following simultaneous bilateral segmental resection.

resections are of course still receiving chemotherapy. The one positive gastric developed in the staged series where only two weeks of streptomycin and isonicotinic acid hydrazide had constituted the preoperative preparation. It now appears that the persistent disease in this patient is from an area corresponding to the superior segment of the left lower lobe where a wedge resection was performed. Incomplete resection could well play a part in the gastric relapse.

COMMENT

The introduction of simultaneous bilateral segmental resection for pulmonary tuberculosis should prove of definite value in selected cases. With the anterior two-team approach there are few technical hazards. One case manifested a severe air leak on the right immediately postoperatively (Patient 2 of the simultaneous group). By dividing the suction so that a pressure of 40 cm. of water was available to the right thoracic cavity, while the left was maintained on 15 to 20 cm., the residual lung expanded to its full capacity. None of the three simultaneous cases developed any breathing difficulty. The union of the sternal joint must be firm to prevent excessive pain which could embarrass respiration as well as compromise the cough reflex. Bronchoscopy and planigrams in selecting simultaneous bilateral cases are very important. Finally, as in all excisional treatment for pulmonary tuberculosis, the operation indicated by the disease must be performed. This requires careful preoperative evaluation. Wedge resections alone seldom constitute an adequate procedure. In such instances as these, it is doubtful that the surgery was justified; if it was justified, spread or persistent disease is a real possibility.

SUMMARY AND CONCLUSIONS

1. Bilateral pulmonary resection for tuberculosis is a safe and practical procedure.

2. Twelve patients were subjected to staged bilateral resections without any deaths. One gastric relapse has occurred; otherwise, early results are favorable.

3. Simultaneous bilateral segmental resection is suggested for the relatively good-risk patient who requires bilateral surgery. The advantages of the simultaneous approach are discussed.

4. Operations in three simultaneous bilateral cases have been performed without serious incident. An anterior, transverse sternotomy incision is used with the two-team operating system.

ADDENDUM

Since this paper was submitted for publication, five additional simultaneous bilateral resections have been performed without mortality. In one case, right upper lobectomy was combined with an apicoposterior segmental excision on the left to substantiate the contention that resection need not be limited to the segmental level.

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DICED CARTILAGE FOR CHEST WALL DEFECTS

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THE resection of two or more segments of ribs, together with periosteum and intercostal muscles, deprives that portion of the thoracic wall of its normal stability and results in herniation of the lung and paradoxical motion during respiration. The severity of these effects depends on the size of the defect and the amount and thickness of the structures overlying it. Underneath the scapula or the trapezius and rhomboidei muscles, the effects are minimal, but when ribs are removed from the lateral chest wall, where there is the least amount of muscle tissue, paradoxical motion is most severe. For over sixty years, various methods have been used to restore the stability and protection of the chest by securing a firm closure of chest wall defects. The introduction of the use of antibiotics and tantalum stimulated greater interest and additional attempts to solve this problem.

L. Mayer,[15] in 1941, reported the successful use of a free graft of fascia lata to close a large chest wall defect following the removal of a chondrosarcoma of a rib. Watson and James[21] reported similar success in 1947. Davis and associates[9] and Campbell,[8] in 1950, mobilized and transplanted the latissimus dorsi over a free fascia lata graft to give added support to close a large chest wall defect. Effler and Blades,[10] in 1946, and Beardsley,[2] in 1950, reported the unsuccessful use of rigid tantalum plates. The plates had to be removed because of persistent drainage and infection. Uniformly successful closures were obtained with the use of tantalum gauze by Beardsley and Cavanaugh,[3] Morrow,[16] and Ada and Hevenor.[1] Maier[13] mobilized and used the breast to protect anterior chest wall defects. Pickrell and associates[18] utilized skin and muscle pedicle flaps, and Priolean[19] used full-thickness skin flaps to cover the defects. Mauer and Blades[14] mobilized segments of adjoining periosteum and split segments of ribs to cover small and medium-sized defects. Kinsella and associates[12] divided a tibial graft into segments and Bisgard and associates[4] used mortised split rib grafts to overcome sternal defects following the resection of sternal tumors. Hardin and associates[11] reported the successful use of preformed Lucite plates to cover large chest wall defects. Blades and Paul,[5] however, reached the conclusion: "The use of tantalum plates and other foreign bodies is unnecessary and unsatisfactory" and transplants of fascia lata "are rarely necessary."

The method described by Mauer and Blades[14] appears to be the best for the small chest wall defects because autogenous adjoining tissue is utilized to form a bony support. The only disadvantage of fascia lata or tibal grafts is that an additional operative procedure is necessary. Thus far, it appears that

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the most popular method for the closure of large chest wall defects is by the use of tantalum gauze. The desire to obtain a strong rigid cover over an exposed heart and the curved anterolateral portion of the chest wall provoked the idea in the authors to use diced cartilage between two layers of tantalum gauze.

Peer[17] was the first to utilize diced cartilage to cover skull defects in 1943. Subsequently, its use has been extended to restore shape in bony deficiencies of the face, for construction of ears, to form an articular surface of cartilage in ankylosed hips, to reinforce fascia in the repair of hernias, and to cover bony defects in spina bifida.

Diced cartilage grafts, as the name suggests, consist of numerous small segments of cartilage which can be packed or molded into any desired contour, like wet grains of sand. Diced cartilage can be quickly prepared by putting through an ordinary meat grinder pieces of cartilage preferably obtained from young subjects who have met death by accident. It may be stored indefinitely in a freezer "bank" until it is used. Microscopic examination of the diced cartilage grafts following transplantation at various intervals up to four years has been made. The sections demonstrated that the spaces between the small cartilage segments were occupied first by blood and later by ingrowing connective tissue accompanied by numerous small blood vessels. Each small cartilage graft rested against adjoining cartilage at some points with a thin layer of connective tissue between, thus preventing contracture of the cartilage mass. The collective bulk of the diced cartilage grafts was increased by the addition of the numerous small spaces between the pieces of cartilage, and for this reason, the diced cartilage grafts actually filled a larger space than the solid rib cartilage from which they were cut. Autogenous diced cartilage grafts had living chondrocytes, normal-appearing matrix, with occasional bone formation in or outside the grafts and a general absence of invasion and absorption. Preserved diced cartilage grafts showed definite invasion and absorption in some grafts, whereas in others the matrix, with dead cells, appeared to resist invasion quite well. The tendency to bone formation was more pronounced in the preserved cartilage than in the autogenous cartilage. Connective tissue and blood vessels grow through the openings of the mesh and join the diced cartilage grafts into a solid structure. The ingrowing host of fibroblasts, together with collagenous fibers and cement substance, provides a strong binding wall, in which the individual cartilage segments are firmly held together to protect the vital structures beneath.

CASE REPORT

B. McG., a white boy aged 15, on Aug. 1, 1942, was struck by an ice truck and sustained a crushing injury to his anterior chest wall with multiple rib fractures. He was treated conservatively, and discharged from the hospital with a marked anterior chest wall deformity which consisted mainly of a depression involving the left fifth, sixth, seventh, and eighth costal cartilages and elevation and angulation of the right fourth, fifth, and sixth costal cartilages. In 1948, a plastic operation was performed at Mt. Sinai Hospital in New York to correct the deformity by resecting the deformed cartilages. As a result of an infection, a draining sinus persisted until October, 1950, when he was admitted to the New York Hospital. The draining sinus was excised, and a chronic empyema cavity was drained and a

thoracoplasty performed. Several operations were necessary to clear up the infection before healing took place in July, 1951. At this time, he had a right parasternal defect which involved the loss of about 4 cm. of the right fourth, fifth, and sixth costal cartilages, and a very large defect which involved the loss of the anterior fifth, sixth, seventh, and eighth ribs and cartilages. The heart was covered only by the pericardium and a wide paper-thin scar. The scar extended horizontally from the sternum, where it was widest, measured 8 cm., gradually tapered down to a linear scar in the left axilla, and continued around the angle of the scapula. The entire anterior surface of the heart was covered by this thin scar so that the heart could be grasped with the fingers (Fig. 1). Paradoxical motion was marked over the defects, and the heart action was very apparent.

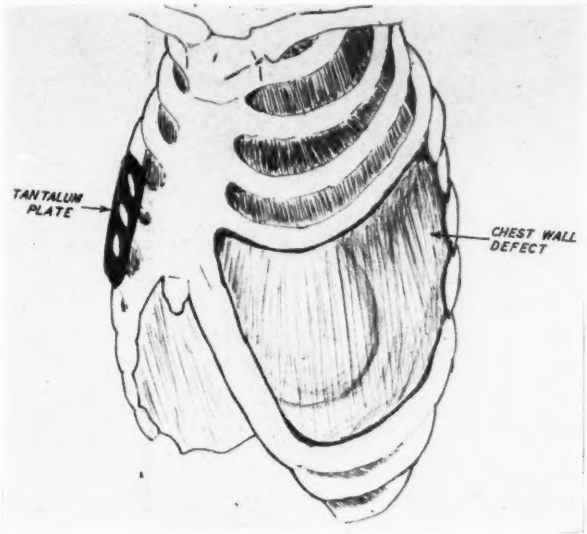

Fig. 1.—Thoracic cage showing the tantalum plate covering the bony defect on the right anterior chest wall following the first operation, and the large chest wall defect on the left, through which the heart could be easily felt.

He was repeatedly refused employment because of the large chest wall defect and exposure of his heart. He became a client of the New Jersey Rehabilitation Commission because of his employment handicap. He was admitted to the St. Barnabas Hospital in Newark for plastic closure of the chest wall defects. It was decided first to repair the right side by means of a tantalum plate fashioned to an oval shape to barely overlap the edges of the defect. Three horizontal segments of the plate were cut out to correspond to the missing intercostal spaces. Small drill holes were made along the edge of the plate for steel sutures. On June 20, 1952, the scar was excised down to the pleura. The pectoralis muscle and fascia were mobilized and the ends of the ribs and edge of the sternum were exposed. Suture holes were made in the ends of the ribs and sternum with a towel clamp. The tantalum plate was firmly anchored with steel sutures over the defect (Fig. 1). The pectoralis muscle and fascia were sutured over the plate with interrupted chromic catgut sutures and the skin was closed with silk. The wound was not drained, but it was necessary on two occasions to aspirate a small amount of accummulated bloody fluid. The wound healed cleanly.

The patient was readmitted to the hospital on Nov. 12, 1952. The tantalum plate on the right side had proved very satisfactory, but this method was not considered for the

left side because of the extent of the defect and mobility of this part of the chest. The maximum protection and a certain degree of flexibility were desired and it was felt that a combination of diced cartilage and tantalum gauze would provide these advantages. On November 17, the entire horizontal scar on the left chest, including the wide paper-thin scar which was attached to the anterior pericardium overlying the major portion of the cardiac area, was excised. The scar was dissected off the anterior pericardium and the lingular portion of the upper lobe of the left lung. The overlying thickened pleura was decorticated and the atelectatic lingula freed and re-expanded. The incision was developed by reflecting the skin, subcutaneous tissue, and muscles off the fourth and ninth cartilages and ribs. The fifth, sixth, seventh, and eighth cartilages and anterior ribs were missing from the sternum to the left axilla as a result of the previous operations (Fig. 1). The fourth and

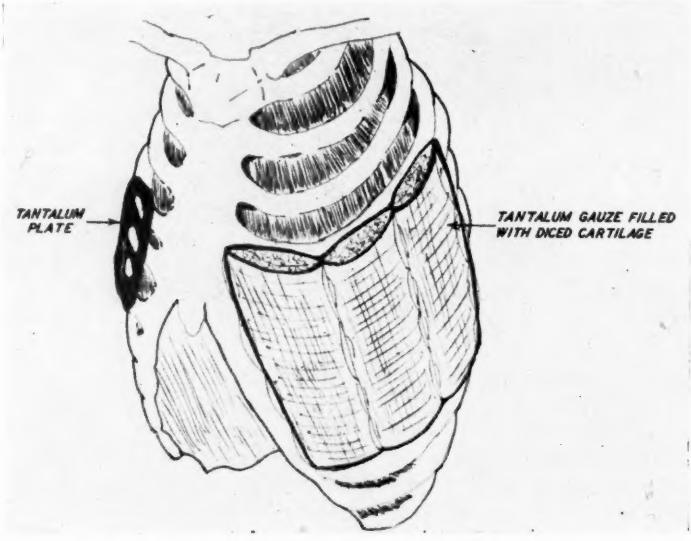

Fig. 2.—Shows the large trilocular tantalum gauze, filled with diced cartilage, sutured into position and covering the chest wall defect.

ninth ribs were divided horizontally. The medial end of the ninth rib was sutured to the fourth rib in the manner of Mauer and Blades,[14] so that they lay diagonally across the cardiac area and served as struts and supports for the tantalum mesh. A sheet of tantalum mesh was folded in half and the sides were sutured together with interrupted wire sutures. Two similar vertical rows of interrupted wire sutures through both layers of the mesh formed three compartments. These were filled with diced "bank" cartilage. The cartilage-filled tantalum mesh was placed on the chest wall defect and molded into shape. It was anchored into position with interrupted wire sutures to the fourth, and ninth ribs, and sternum. (See Fig. 2.) The graft was covered with as much muscle and fascia as could be mobilized. The skin was closed with interrupted silk sutures. The wound was drained with a catheter to which continuous suction was connected. Drainage was moderate the first two days, and the catheter was removed on the fourth day. Sutures were removed on the seventh day. He was discharged on the twenty-first postoperative day with a healed wound except for a small granulating area at the medial end. He became fully employed in the early part of 1953, and has remained so since.

In spite of the maximum efforts to elevate the sternum, following a chondrosternoplasty operation for congenital chondrosternal depression or funnel

chest, there often remains a varying amount of depression in the gladiolar-xiphoid area. In spite of the physiologic relief in elevating the sternum from the compressed heart, patients are so expectant of having a normal-shaped chest that they are keenly disappointed with any degree of depression. In every chondrosternoplasty, there remains always a considerable excess amount of pieces of resected costal cartilage. Brodkin[6, 7] has now modified his technique of chondrosternoplasty by dicing these excess pieces of cartilage, deflecting the outer periosteum of the sternum, and filling the space with the diced cartilages. The result has been most gratifying, as can be seen in the illustrations. (Figs. 3 and 4.) Traction with steel wire through the lower end of the sternum was used in this patient.

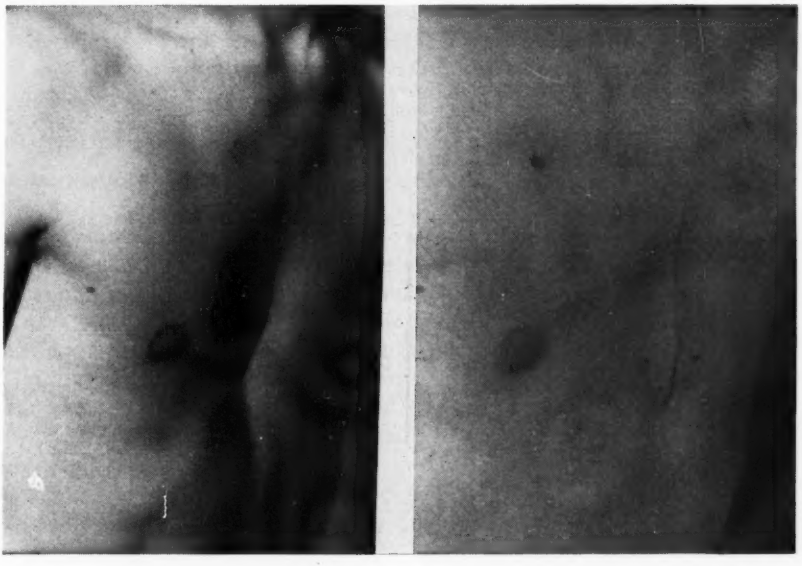

Fig. 3. Fig. 4.

Fig. 3.—R. C., aged 16, with severe congenital chondrosternal depression (funnel chest). The depression held 140 ml. of fluid.

Fig. 4.—Postoperative view following a chondrosternoplasty in which autogenous diced cartilage was used to fill the residual depression.

Urban and Baker[20] create a chest wall defect after resecting the second to the sixth costal cartilages to remove the internal mammary vessels and lymph node chain during a radical mastectomy for carcinoma in the medial half of the breast. They repair the defect with a free fascia lata graft. To reduce the time of the operation, avoid an additional operative procedure, and restore a solid repair of the chest wall, the defect is repaired with a double layer of tantalum gauze between which diced cartilage obtained from the resected cartilage is molded and placed. This can be done by the assistant while the surgeon is dissecting the axilla.

SUMMARY

An additional method is suggested to repair chest wall defects. Diced cartilage has proved to be very successful as a graft in places where solid support is necessary. It is a very practical replacement, because autogenous cartilage is available, and, when necessary, stored "bank" diced cartilage will prove equally efficacious. In large chest wall defects, particularly when protection is desired as in the precordial area, a solid wall will be secured with stored diced cartilage. In patients undergoing a chondrosternoplasty for congenital chondrosternal depressions (funnel chest), a better cosmetic result will be obtained. In the repair of the defect resulting from the resections of the costal cartilages in radical mastectomy, a solid replacement may be obtained by using autogenous diced cartilage and tantalum gauze.

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A STUDY OF THE EFFECT OF PARA-AMINOSALICYLIC ACID ON BLOOD COAGULATION AND PROTHROMBIN CONCENTRATION ON PATIENTS WITH TUBERCULOSIS

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THIS study was undertaken to determine whether para-aminosalicylic acid (PAS) as it is ordinarily used in tuberculosis therapy has either a prothrombinopenic effect similar to its analogue, sodium salicylate,[5, 7-9, 15] or an anticoagulant action caused by the release of heparin from the mast cells as suggested by Soragni [16, 17]; and finally, whether it might in some instances be responsible for serious hemorrhage during the course of thoracic surgery.

LITERATURE

Norvit and Carstensen recently reported, after an extensive study of 400 patients, that PAS had no effect on prothrombin concentration, and was never responsible for an increased hemorrhagic tendency.[10] Croizat, on the other hand, reported the case of a 20-year-old white woman with tuberculosis who developed a spontaneous hemorrhage which he attributed to PAS. Her prothrombin concentration was 42 per cent, and, according to this author, was the only reasonable explanation for the hemorrhage.[2] Gordin,[4] Forsgren,[3] Tornaky,[19] Swanson,[18] Barclay,[1] Postiglione,[12] Phillips,[11] and Lynch[6] found that PAS had a small but definite effect upon prothrombin concentration. Several of these investigators expressed the opinion that the effect of PAS on prothrombin concentration was accentuated in patients with liver disease. Soragni reported that although the prothrombin concentration rarely dropped below 60 per cent in adults treated with PAS, children were affected more profoundly. The decrease in prothrombin concentration was independent of the dose given, was not progressive, but showed individual variation, and also varied from day to day in the same individual. He found further that the effect was rapidly reversed by administering vitamin K and discontinuing the PAS. He conjectured without experimental evidence that in addition to causing hypoprothrombinemia, PAS might have a further anticoagulant action by releasing heparin from the mast cells.[16, 17]

METHODS AND MATERIALS

The present study consisted of making multiple prothrombin time determinations on eighty-one patients with tuberculosis who were undergoing PAS therapy, with a follow-up after PAS had been discontinued on those patients who were found to have developed hypoprothrombinemia. These patients had been receiving 12 Gm. of liquid sodium PAS daily in divided doses for varying periods of time from ten days to over one year. Prothrombin time determinations

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were also made on thirty control patients who had tuberculosis, but had not been on PAS. Second, coagulation time studies were performed on a group of thirty patients with tuberculosis before, and two hours after, a 6 Gm. dose of liquid PAS had been given. Blood levels of PAS were also determined. Last, an attempt was made to determine whether or not there were any instances of abnormal bleeding at surgery or in the immediate postoperative period on the thoracic surgical service at this hospital during the twenty-three-month period from Jan. 1, 1951, to Dec. 1, 1952, that might have been caused by preoperative PAS therapy. During this interval, there had been a total of 288 major thoracic operations performed, including 200 lung resections of various types, forty-two thoracoplasties, seventeen decortications, and twenty-nine miscellaneous procedures. Over 90 per cent of these operations were preceded by a course of PAS and streptomycin.

The prothrombin time determinations were done by the Quick one-stage prothrombin method, using acetone-dehydrated rabbit brain as the source of thromboplastin.[14] Multiple determinations were made on each patient on different days and mean values were used for comparisons. The prothrombin times of normal nontuberculous controls using this method varied from twelve to thirteen seconds.

A modification of the Lee-White method was employed for the coagulation time studies.[13, 14] These were performed as carefully as possible with due attention to the use of scrupulously clean equipment, atraumatic venepunctures, and at controlled temperatures (37° C.). To assure the fact that the PAS administered to the patient had been absorbed, a blood level was obtained at the time of the second coagulation time test, approximately two hours after the drug had been taken. For this determination, a modification of the Bratton-Marshal sulfonamide assay method was employed.[20]

RESULTS

The results of these various determinations can be summarized as follows: (1) PAS was found to have no demonstrable effect on the coagulation time of blood at concentrations of the drug that were found to average 11.28 mg. per cent. (2) The drug had a slight effect on prothrombin concentration in eleven of the eighty-one patients who were being treated with it. In these eleven patients, prothrombin time prolongations varied from 2.8 seconds (55 per cent concentration) to 5.6 seconds (32 per cent concentration). In follow-up studies performed on ten of these patients after PAS had been discontinued, all but three had returned to control or very near control values. Of the three patients who failed to return to normal, two had poor liver function but the third had no demonstrable abnormalities, and the etiology of his persistent hypoprothrombinemia was not established.

The effect of discontinuing PAS in the ten patients with prothrombin prolongations is depicted in Fig. 1.

The effect of PAS on the prothrombin time of one of the patients (Patient 5) is shown in Fig. 2. (3) In the third and final portion of this study, it was

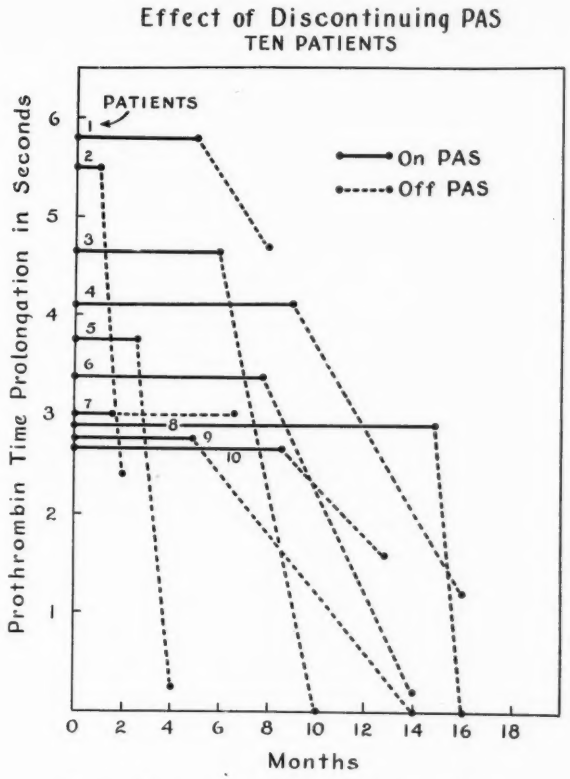

Fig. 1.—Effect of discontinuing PAS: ten patients.

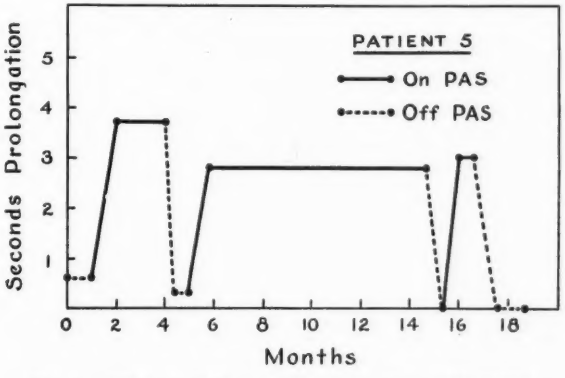

Fig. 2.—Effect of PAS on prothrombin time prolongation.

found that there were no instances of excessive bleeding due to abnormal blood coagulation in the entire group of 288 major thoracic operations during the period under study. This observation must be qualified by stating that during the latter half of the period, vitamin K was given to some of the patients who were found to have hypoprothrombinemia. Whether any of these would have bled without vitamin K is, of course, not known. The absence of abnormal bleeding in this group of surgical operations confirms the impression gained from these experimental data and from most of the literature that the hypoprothrombinemia caused by PAS in a total daily dose of 12 Gm. is not of sufficient severity to cause either spontaneous hemorrhage or abnormal hemostasis during the course of surgery.

These observations, however, do not preclude the possibility of severe hypoprothrombinemia developing in a patient who for one reason or another might be unusually sensitive to PAS.

Because of this possibility, it would be advisable to check the prothrombin concentration of all patients who are receiving PAS one week or so prior to surgery.

SUMMARY

An attempt has been made to evaluate the effect of PAS on the prothrombin concentration and coagulation time of blood and, further, to ascertain whether or not this effect is of clinical importance. PAS was found to have no discernible effect upon the coagulation time of thirty patients studied. A moderate prolongation of the prothrombin time over control values was found in eleven of eighty-one patients studied; these prolongations varied from 2.8 seconds to 5.8 seconds. All but three of these prolonged prothrombin times returned to normal values after PAS had been discontinued.

There was no clinically evident adverse effect upon blood coagulation, because in 288 surgical procedures, 90 per cent of which were preceded by PAS, there were no instances of abnormal hemostasis at the time of surgery or in the immediate postoperative period.

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Announcements

THE AMERICAN ASSOCIATION FOR THORACIC SURGERY

Notice of Annual Meeting

The Thirty-fifth Annual Meeting of the American Association for Thoracic Surgery will be held April 24, 25, and 26, 1955, in Atlantic City, New Jersey. Headquarters will be at the Chalfonte-Haddon Hall.

Requests for Hotel Reservations

These should be mailed directly to Miss Doris Sears, Convention Manager, Chalfonte-Haddon Hall, Atlantic City, New Jersey. Please mention this Association, the type of accommodation desired, date and approximate hour of arrival. If accommodations are desired elsewhere in Atlantic City, please communicate directly with the hotel of your choice.

Thoracic Surgical Forum

By vote of the membership, the Forum is to be continued for the presentation of short papers on current thoracic research, experimental problems, anatomical studies and modified or new surgical techniques. The inclusion of any considerable amount of clinical material in these reports makes the choice more difficult and the Program Committee reserves the right to reject such papers or to suggest their transferral to the regular program. Papers at this session will be limited sharply to ten minutes, including the showing of lantern slides or other illustrative material. At the discretion of the President, three minutes may be allowed as the maximum discussion for each paper.

These papers will be published together in one issue in the JOURNAL OF THORACIC SURGERY. Maximum length: 3,000 words without illustrations. If illustrated, an appropriate number of words must be subtracted for each cut used.

Abstracts for Papers

Abstracts for the presentation of papers at the 1955 meeting, including papers before the Forum, must be received on or before Dec. 15, 1954; otherwise they will not be considered by the Program Committee.

Abstracts should be labeled "For Thoracic Surgical Forum," or "For Regular Program."

Five, repeat, five copies of each abstract must be sent to the Secretary of the Association, Dr. Paul C. Samson, 3959 Happy Valley Road, Lafayette, California. They should not be sent to the Editor.

The abstracts should contain from 200 to 250 words and accurately reflect the contents of the completed paper.

Applications for Membership

Applications for Associate Membership in the Association must be received by the Membership Committee not later than Dec. 15, 1954; otherwise, the applications will be deferred for consideration until 1956.

Applicants must be sponsored by three Active or Senior members of the Association. In addition to the signatures on the application form, the sponsors must forward a *separate* letter concerning the applicant directly to Dr. Lyman A. Brewer, III, Chairman of the Membership Committee, 2010 Wilshire Blvd., Los Angeles 5, Calif.

Completed application forms and the sponsoring letters *independently* sent by the three sponsors must be received by Dr. Brewer not later than Dec. 15, 1954, to be considered for action in Atlantic City.

Sponsors are reminded that *new* letters of recommendation must be forwarded to the Membership Committee should their applicants be held over for reconsideration a second year.